Hypodontia and Self-Esteem

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Synopsis

To provide treatment of "the good kind", as described by a subject with hypodontia in the present study, the dental profession must, not only understand the characteristics and complexities of hypodontia, but also, the complexities of those being treated.

The majority of subjects attending the Hypodontia Clinic in Glasgow Dental Hospital and School are teenagers. The adolescent years are generally regarded as a volatile time for those individuals and their parents/guardians. Adolescents are concerned with creating the "right image", physical as well as facial, and are subjected to the influences of parents, peers and the media. Appearance is considered to be paramount for being, successful in many aspects of life and being popular. The presence and the appearance of teeth contribute to facial aesthetics. Appearance, and therefore, self-image and how subjects perceive themselves, is contributory to how subjects feel about themselves. Having self-esteem is about feeling good about ones' self and is important in wellbeing. Therefore, for those with hypodontia, appearance of their teeth is an issue, but to what degree and to what effect. To date, there is no research regarding the degree to which hypodontia affects those subjects, especially in respect of self-esteem.

The clinical effects of hypodontia can be profound requiring the expertise of a multidisciplinary team for corrective treatment. There are many studies which have investigated the prevalence of hypodontia, the most common tooth, and the genetics of hypodontia within a family. The genetics and their interactions associated with hypodontia is not only fascinating but is still unfolding, with

many of the investigations being undertaken with mice. However, research into specific patterns of congenitally missing teeth in familial hypodontia is scanty.

The aim of this research was to endeavour to characterise those subjects referred to the Hypodontia Clinic at Glasgow Dental Hospital and School, with special reference to the patterns of congenitally missing teeth and their associated genes. Furthermore, the degree to which hypodontia affects adolescent subjects was investigated, with specific regard to self-esteem.

In order to characterise those subjects who attended the Hypodontia Clinic at Glasgow Dental Hospital and School, a hypodontia database was constructed to record the findings of their clinical assessment.

To record how adolescent subjects with hypodontia felt about their teeth and their treatment needs, participants of 13-18 years of age, were invited to complete the Hosey-Bradnock questionnaire. This is a piloted but not published questionnaire.

To evaluate the self-esteem of adolescent subjects with hypodontia, participants aged 13-18 years, were invited to complete the Culture-Free Self-Esteem Inventory, which has been validated and published. A control group of subjects without hypodontia, also aged 13-18 years, attending the Community Dental Clinic in Clydebank, were invited to complete only the Culture-Free Self-Esteem Inventory.

Seventy one subjects were recruited for inclusion in the hypodontia database. The characterisation of the participants mirrored expected trends in respect of the most common missing teeth, gender ratio, presenting complaint and treatment. There was, however, an unexpected and unexplained preponderance of missing maxillary canines. The patterns of sixty seven of these subjects were able to be interpreted, in respect of the gene markers of Msx 1 and Pax 9. Another possible gene marker, transforming growth factor alpha (TGFA), was also included, in the light of the high involvement of missing maxillary lateral incisors in the patterns observed. The results showed that in this sample that, only three subjects showed patterns of missing teeth which correlated to the specific pattern of congenitally missing teeth identified for Msx 1 and Pax 9. The majority of the patterns were thought to either be the result of mutations of Msx 1, Pax 9 or an unknown gene or, interactions between Msx 1, Pax 9, TGFA and an unknown gene or genes, in combinations. Despite, the selection criteria excluding those with syndromes, there was also the possibility that some subjects within this sample were either, carrier status for or, had undiagnosed syndromes. Ectodermal dysplasia was thought of, as the prime suspect. Only genetic analysis of the sample would eliminate or confirm this suspicion.

The Hosey-Bradnock questionnaire was completed by 28 subjects with hypodontia. Of these, 20 (71%), complained about the appearance of their teeth, the vast majority highlighting spaces and gaps. Twenty one of this sample had one or more upper anterior teeth missing. Furthermore, 18 (64%) subjects "hated" or "disliked" their teeth.

These aforementioned 28 subjects with hypodontia and 30 controls accepted the invitation to participate in the controlled study to evaluate the self-esteem of adolescents with hypodontia. The Culture-Free Self-Esteem Inventory had an inbuilt "lie detector" within it, so that the measure of self-esteem, the Global Self-Esteem Quotient (GSEQ) of only truthful subjects was analysed statistically. The Mann-Whitney test showed that the hypodontia subjects did not have a lowered self-esteem ($p=0.8$). This was a gratifying, but surprising result, given that the vast majority of the adolescent hypodontia subjects complained about appearance of their teeth. These subjects and their parents obviously deemed teeth and their appearance to be a priority or they would not attend the appointments or embark on often complex treatment plans. This finding of the lack of lowered self-esteem, may be attributed to the fact that these subjects realised there was a problem but they, and their parents/guardians had coped with it by accessing the services for corrective treatment. Furthermore, positive support from parents can only promote positive esteem and wellbeing in their children.

Dedication

To my parents, for fostering in me, by example, the love of learning, the enquiring mind and the ethos of hard work.

To my husband, Michael and my daughter, Emma, for their understanding, love and support.

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Declaration

This thesis represents the original work of the author

The work of the following people prior to this included:

- Construction and piloting of the Hosey-Bradnock questionnaire by Dr M.T. Hosey and Dr G Bradnock (unpublished).
- The construction and validation of the Culture-Free Self-Esteem Inventory by J Battle (2002).

Rosemary Dunlop Broad BDS

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Chapter 1 Literature Review.

1.1 Introduction.

Every human is individual and unique. Their differing appearance and characteristics are governed by their genetic make-up and environmental influences. Sometimes, because of up-sets within the genetic make-up or unfavourable environmental influences, variations arise outwith the "normal" resulting in anomalies.

Teeth show many variations from the "normal" in respect of size, shape, structure, number and position. One of the most common dental anomalies is that involving a lack of the normal number of adult teeth:-hypodontia (On-line Mendelian Inheritance in Man http;//www.ncbi.nlm.nih.gov/OMIM). This is approximately six times more common as extra teeth (supernumary) (Thilander and Myrberg 1973).

1.2 The normal number of teeth.

In adults, the normal number of teeth is twenty-eight permanent teeth with variable presence of up to four third permanent molars. In children, the normal number of primary (baby) teeth is twenty. Both dentitions are equally divided between the maxilla and the mandible.

1.3 Definition of hypodontia.

Some people may fail to develop the normal number of teeth in either the primary or permanent dentitions or in both. A tooth that fails to develop is considered to be congenitally missing or absent (Brekhus et al 1944).

Jorgenson (1980), stated that *hypodontia* is best defined as agenesis (non development) of one or more teeth. He suggested that the term congenital "absence" is less clear because teeth are rarely evident at birth and many do not begin to develop until after birth. Despite this, most of the literature generally refers to hypodontia as the congenital absence of one or a "few" teeth. *Oligodontia* is the term used when "many" teeth are congenitally absent. These terms can be used for both the primary and the permanent dentition. The terminology of hypodontia and oligodontia is vague; "few" and "many" are seldom given a numerical value. Where, Schalk-van der Weide et al (1994) defined oligodontia as the congenital absence excluding third molars of six or more teeth, Øgaard and Krogstad (1995) considered it to be ten or more.

In severe cases, the complete absence of teeth either in the primary dentition, the permanent dentition or both is referred to as *anodontia*.

1.4 Prevalence of hypodontia.

Prevalence studies vary considerably in their findings. These are unlikely to be attributable to true variations but a contrast in sampling techniques, examination methods and diagnosis (Brook 1975, Jorgenson 1980).

1.4.1 Prevalence of hypodontia in the primary dentition.

Hypodontia in the primary dentition is infrequent and studies suggest a range of 0.1-0.9 per cent (Menczer 1955, Saito 1959, Grahnén and Granath 1961, Ravn 1971, Brook 1974, Järvinen and Lehtinen 1981, Magnusson 1984). The aforementioned author found in his study of 741 English children aged three to five years old, that the prevalence was 0.3 per cent. Table 1.1 overleaf shows the prevalence studies from different countries and samples.

Hypodontia in the primary dentition generally affects the maxillary lateral incisor (Clayton 1956, Grahnén and Granath 1961, Ravn 1971, Järvinen and Lehtinen 1981, Magnusson 1984, Daugaard-Jensen et al 1997a). The next most common missing primary tooth was the mandibular lateral incisor (Ravn 1971, Järvinen and Lehtinen 1981 and Daugaard-Jensen et al 1997a).

Table 1.1 Prevalence of hypodontia of the primary dentition.

Author(s)	Country	Sample size	Prevalence
Menczer (1955)	U.S.A.	2,209 Pre-school dental clinic (2-6 years)	0.1%
Saito (1959)	Japan	7,589 Kindergarten and nursery	0.2%
Grahnén and Granath (1961)	Sweden	1,173 Department of Pedodontics Royal Dental Hospital, Palmo (3-5 years)	0.4%
Ravn (1971)	Denmark	4,564 Infant dental service (3-3.5 years)	0.6%
Brook (1974)	England	741 Nursery and infant schools (3-5 years)	0.3%

Järvinen and Lehtinen (1981)	Finland	1,141 Child welfare centres (3-4 years)	0.9%
Magnusson (1984)	Iceland	927 Two health centres three kindergartens seven pre-school classes (dental stage DS 02/primary teeth fully erupted)	0.5%

1.4.2 The prevalence of hypodontia in the permanent dentition.

Hypodontia is more commonly seen in the permanent dentition and there is general acceptance of prevalence in the range of 3.5-6.5 per cent, excluding the third molars. Brook (1974) in his study of 1,115 English schoolchildren, aged 11-14 years old, found a prevalence of 4.4 per cent. Grahnén (1956) and Thilander and Myrberg (1973) reported the prevalence of third molar hypodontia as 25 and 27 per cent respectively.

Hypodontia in the permanent dentition generally follows in the descending order of third molars, mandibular second premolars, the maxillary second premolars and maxillary lateral incisor (Grahnén 1956). Table 1.2 overleaf shows prevalence studies in different countries including samples sizes and the most common missing permanent tooth, excluding third molars.

Table 1.2 Prevalence of hypodontia of the permanent dentition including the most common missing tooth.

Author (s)	Country	Sample size	Prevalence	Most common missing permanent tooth
Dolder (1937)	Switzerland	10,000 (Schoolchildren)	3.4%	Mandibular second premolar
Werther and Rothenberg (1939)	U.S.A	1,000 (Dental School Clinic)	2.3%	Maxillary lateral incisor
Byrd (1943)	U.S.A	2,835 (Children 4-14yrs)	2.9%	Mandibular second premolar
Brekhus et al (1944)	U.S.A.	11,487 (Dental Clinic)	1.5% (including third molars)	Maxillary lateral incisor
Grahnén (1956)	Sweden	1,006 (Schoolchildren 11-14 years)	6.1%	Mandibular second premolar
Sabes and Bartholdi (1962)	U.S.A	40,204 (Hospital records)	0.39%	Mandibular second premolar
Niswander and Sujaku (1963)	Japan	4,150 (Schoolchildren)	6.6%	Mandibular lateral incisor

Rose (1966)	England	6,000 (Orthodontic subjects)	4.3%	Mandibular second premolar
Muller et al (1970)	U.S.A.	13,469 Caucasian 1,481 African-American	3.5% 3.7%	Maxillary lateral incisor
Egermark-Eriksson and Lind (1971)	Sweden	3,327 (Schoolchildren)	6.2%	Not reported
Thilander and Myrberg (1973)	Sweden	5,459 (Schoolchildren)	6.1%	Mandibular second premolar
Hunstadbraten (1973)	Norway	1,295 (Schoolchildren)	10.1%	Mandibular second premolar
Brook (1974)	England	1,115 (Schoolchildren 11-14 years)	4.4%	Not reported
Magnusson (1977)	Iceland	1,122 (6 subjects excluded) (Schoolchildren 8-16 years)	7.9%	Mandibular second premolar
Rolling (1980)	Denmark	3325 (Schoolchildren)	7.8%	Mandibular second premolar
Aasheim and Ögaard (1993)	Norway	1953 (Orthodontic Department)	6.5%	Mandibular second premolar

1.4.3 The effect of gender on the prevalence of hypodontia.

In the primary dentition, there is no significant gender difference between males and females (Brook 1974, Ravn 1971, Clayton 1956, Järvinen and Lehtinen 1981, Daugaard-Jenson et al 1997b).

In the permanent dentition, females are affected more frequently by hypodontia (Brekhus et al 1944, Rose 1966, Brook 1974, Davis and Darvell 1993). However, many studies have reported that there is no gender difference (Dolder 1937, Werther and Rothenberg 1939, Rolling 1980, Aasheim and Ögaard 1993).

In his study of English school children, Brook (1974) reported that males had a higher prevalence of supernumeraries and invaginated teeth (a type of malformed tooth) compared to females, whilst females had a higher prevalence of hypodontia and microdontia (small shaped teeth). Similarly, Niswander and Sujaku (1963), in his study of Japanese children, found the gender differences highly significant, with females again having more congenitally missing teeth and the males having more supernumeraries. These confirmed the report by Brekhus et al (1944) who, in their study of 11,487 subjects, found that, of the 195 presenting with dental anomalies, 184 presented with hypodontia and microdontia, and that 60% of those with hypodontia were female.

Rose (1966) and Dolder (1937) reported that where only upper lateral incisors were missing, females outnumbered males. Furthermore, Magnusson (1977) found that malformed teeth accounted for 2.2% of the 1,122 children in his study

and not only did the females have a higher prevalence compared to the males but that hypodontia was found in one-third of the girls with malformed teeth.

Conversely, Rolling (1980) found that more males than females had one congenitally missing tooth, however, girls were more affected where multiple teeth were missing. Dolder (1937) found that upper premolars were more commonly missing in the male.

Rose (1966) in his study of 6,000 orthodontic subjects of which females accounted for 60%, found hypodontia in approximately 68% of females. This study, however, found females marginally more predisposed to missing upper premolars than males. Table 1.3 overleaf shows the gender distribution of hypodontia in the permanent dentition.

Table 1.3 Gender ratios of hypodontia.

Author (s)	Country	Ratio of affected Males: Females
Grahnén (1956)	Sweden	1: 1.6
Niswander and Sujaku (1963)	Japan	1: 1.4
Rose (1966)	England	1: 1.5
Muller et al (1970)	U.S.A.	1: 1.4 Caucasian 1:1.3 African-American
Egermark-Eriksson and Lind (1971)	Sweden	1: 1.6
Brook (1974)	England	1: 1.8

1.4.4 The effect of race on hypodontia.

Ethnic variation has been reported for hypodontia. Mandibular incisors are frequently absent in Southern Chinese children (Davis and Darvell 1993). In Japanese and Malaysian races, congenital absence also seems to affect the lower incisor region, but particularly the lower lateral incisor (Niswander and Sujaku 1963, Nik-Hussein and Majid 1996). Despite this, interestingly, the frequency of the congenital absence of the upper lateral incisor in the Japanese race was 16.3%, which is close to the 19.6% found by Grahnén (1956) in the Caucasian population (Niswander and Sujaku 1963).

In the study of American subjects by Muller et al (1970), where 13,469 were Caucasian and 1,481 were African-American, the prevalence was 3.5% and 3.7% respectively and the most commonly missing tooth was the upper lateral incisor. In another study, of 1,060 African-American children attending a childcare clinic situated in a hospital gave a prevalence of 7.7% and the most common missing tooth was the mandibular second premolar. However, it is noted that not only was the sample size considerably lower but the socio-economic background of the second study was poor (Salinas and Jorgenson 1974). Garner and Yu (1978) reported that there was a significantly higher incidence of congenitally missing teeth in Caucasians (5.15%) than in African-Americans (2.57%), almost twice as much.

Dolder (1937) reported that the prevalence in Switzerland was 3.4%. Furthermore, differences in race and shape of skull differences were also evident. The short-skulled alpine population showed absence of second premolars to be most common. However, the nordic long-skulled population showed the frequent absence of maxillary lateral incisors and central lower incisors.

The prevalence in the Scandinavian population is reported to be about 5-9%, with the mandibular second premolar most commonly affected. Norwegians account for 6.5% (Aasheim and Ögaard 1993), Danish 7.8% (Rolling 1980), Icelandic 7.9% (Magnusson 1977).

Brook (1974), confined his study to the Caucasian population in England where the prevalence was found to be 4.4% and therefore race differences cannot be compared.

1.5 Relationships to hypodontia.

1.5.1 The relationship of hypodontia in the primary to the permanent dentition.

Hypodontia in the permanent dentition occurs in approximately 75% of individuals with hypodontia in the primary dentition (Grahnén and Granath 1961). Furthermore, this hypodontia in the primary dentition usually corresponds to hypodontia in the same region in the permanent dentition (Ravn 1971, Daugaard-Jensen et al 1997b, Järvinen and Lehtinen 1981, Davis and Darvell 1993, Grahnén and Granath 1961).

1.5.2 The relationship of hypodontia to incisal relationship.

Basdra et al (2001) reported that, in their study of 150 subjects, congenital tooth anomalies were associated with a three fold increase in subjects with a Class II division II incisal relationship compared to normal occurrence rates. In contrast, Class II division I and Class III incisal relationships showed occurrence rates similar to that found in the normal population with respect to subjects with tooth anomalies. However, despite this, the aforementioned authors also reported that descriptive statistics showed a slight trend towards an association between Class III incisal relationship and tooth anomalies.

Nonetheless, Chung et al (2000) reported that in their study of 59 subjects from a sample of 150 subjects with hypodontia that the majority presented with Class III incisal relationship. Selection within the hypodontia sample in the aforementioned study was based on the presence of a lateral cephalometric radiograph. Rose (1966) in his study of 258 subjects with hypodontia from a sample of 6,000 orthodontic subjects, reported that the majority presented with Class I incisal relationship. Furthermore, Rose (1966) compared the incisal relationship of these subjects with hypodontia with a random sample of subjects without hypodontia and reported no statistical association between the two groups. There are sampling differences between the studies of Chung et al (2000) and Rose (1966) and these may account for the differences. Table 1.4, below shows the results in the two studies.

Table 1.4 Incisal relationship in relationship to hypodontia.

Incisal relationship	Rose (1966)	Chung et al (2000)
Class I	57%	31%
Class II division I	29%	10%
Class II division II	5%	25%
Class III	9%	34%

1.5.3 The relationship of hypodontia to skeletal pattern

Chung et al (2000) reported that subjects (n=59) with severe hypodontia showed a trend towards a Class III skeletal relationship. However, Øgaard and Krogstad

(1995) in their study of 44 children with six or more missing teeth compared to controls, found few differences in skeletal discrepancies.

1.5.4 Relationship of hypodontia to other dental anomalies.

Hypodontia is associated with other alterations of dental features. These include a reduction in the size of the remaining teeth, altered tooth morphology, abnormal molar root formation and a delay in the overall rate of dental development (Garn and Lewis 1970, Rune and Sarnas 1974). Other abnormalities of the dentition such as, the malposition of canines, reduced alveolar development, infraocclusion of primary molars, the presence of double primary teeth, rotated premolars and enamel hypoplasia are also associated with hypodontia (Svinhufvud et al 1988, Peck et al 1993, 1994, Kurol and Thilander 1984a and 1984b, Baccetti 1998a, Baccetti 1998b, Lai and Seow 1989, Bjerklin et al 1992, Niswander and Sujaku 1963, Arte et al 2001).

1.6 Hypodontia.

1.6.1 Single missing tooth.

The most common form of hypodontia was of a single missing tooth. This has been reported as being between 50-56 % of subjects presenting with hypodontia (Grahnén 1956, Rose 1966, Aasheim and Ögaard 1993 and Rolling 1980). The upper lateral incisor was the most frequently involved, followed by the lower second premolar and then the upper second premolar in these cases (Muller et al 1970, see Table 1.5 on page 40).

1.6.2 Multiple missing teeth.

Congenital absence of up to two teeth has been reported in 85% of subjects (Grahnén 1956, Rose 1966, Aasheim and Ögaard 1993 and Rolling 1980). Furthermore, Rolling (1980) and Grahnén (1956) also found that hypodontia in the premolar regions and the lateral incisor region accounted for 98% and 82% respectively. Indeed, Rose (1966) reported that, in his study of 6,000 orthodontic subjects, that the list overleaf in descending order of prevalence, accounted for 90% of the total number of missing teeth, excluding third molars:

- Mandibular second premolars.
- Maxillary lateral incisors.
- Maxillary second premolars.
- Mandibular central incisor.

The aforementioned author also reported that premolars accounted for over 60% of the total number of congenitally missing teeth, of which mandibular second premolars accounted for 40.71% and the maxillary second premolars for 19.83%.

Individuals with hypodontia show an increased likelihood of having other types of teeth missing. Both, Ingervall and Hedegaard (1975) and Rolling (1980) also observed that those who had anomalies of upper lateral incisors, second premolars or third molars had a tendency towards congenital absence of other types of teeth. Brekhus et al (1944) reported that 70% of those with hypodontia of premolars also had other teeth missing. Furthermore, Garn and Lewis (1962) reported that hypodontia in other teeth is thirteen times more likely in the presence of

hypodontia of a single third molar. Interestingly, as the incidence of congenitally missing teeth increases, the second premolars are involved more often (Muller 1970, see Table 1.5 below). Generally, an absence of first molars is associated with an absence of other molars (Brekhus 1944).

Table 1.5 The most common missing teeth (CMT) excluding third molars involved in a relation to a number pattern (Muller et al 1970).

	One CMT	Two CMT	Three CMT	Four CMT	Five or more CMT
Maxillary lateral incisor	46.4%	54.2%	22.2%	25.0%	50%
Mandibular second premolars	27.0%	30.0%	83.3%	70.0%	92.9%
Maxillary second premolars	12.1%	11.8%	69.4%	60.0%	92.9%

1.6.3 One type of missing tooth.

Hypodontia has been reported to affect only one type of tooth, with maxillary lateral incisors and second premolars being the most common. Brekhus et al (1944) reported that 50% of 184 cases showing hypodontia lacked one type of tooth (n=92). Of those lacking one type of tooth (n=92), 65.2% were maxillary

lateral incisors, 23.9% second premolars, 4.3% third molars and 6.5% miscellaneous. However, Rose (1966) found the second premolars more common compared to Brekhus et al (1944) although the figures for the maxillary lateral incisors are similar. These comparative results are shown below in Table 1.6.

Table 1.6 Comparison of hypodontia of one type of congenitally missing tooth.

	Brekhus et al (1944)	Rose (1966)
Total of those with hypodontia	184	258
Comparison of the most common "one type of tooth" missing		
Maxillary lateral incisors	60 (32.6%)	67 (25.9%)
Second premolars	22 (11.9%)	149 (57.8%)

1.6.4 Common combinations of missing teeth

Brekhus et al (1944) were the only authors who reported on combinations of missing teeth. These observed common combinations of missing teeth and the three most frequent (in descending order) were:

- one type of tooth
- upper lateral incisor and third molar
- second premolar and third molar.

However, these combinations are strongly linked to the prevalence of the teeth involved, rather than a pattern of certain types of missing teeth. Moreover, this paper was published before the role of genetics in hypodontia was understood. Despite this, it is surprising that there appears to be no further attempt to match the patterns of missing teeth to the likely genetic source.

1.6.5 Abnormality of tooth size and shape in relation to hypodontia.

Hypodontia has been reported to be associated with a reduction in crown size (Garn and Lewis 1970, Baum and Cohen 1971a, 1971b, Rune and Sarnas 1974). Rune and Sarnas (1974) in their study measured the central incisor, the canine and the first molar as these teeth are rarely missing or mal-developed.

Reported crown size reduction is greater in those with multiple missing teeth (Garn and Lewis 1970). Furthermore, Garn and Lewis (1970) reported in their study which included third molars, that there was a relationship with hypodontia and a crown-size reduction gradient in the remaining teeth, with anterior teeth being more severely affected than posterior teeth. The aforementioned authors,

suggested this may be the result of a "field" of size reduction concentrating on the anterior region and diminishing towards the posterior region.

Tooth size appears to have a strong genetic component (Townsend and Brown 1978). A reduction in size and shape of the dentition demonstrates a variation in expression of hypodontia (Grahnén 1956, Alvesalo and Portin 1969). Hypodontia and a reduction in tooth size can occur together either throughout the whole dentition or in some areas of the mouth (Brook 1984).

1.6.5.1 Peg-shaped maxillary laterals.

Magnusson (1977) described peg-shaped teeth as having a mesiodistal width which differed by 2mm or more from its natural size or shape. Manderville (1950), Grahnén (1956), Niswander and Sujaku (1963) and Thilander and Myrberg (1973) reported that when a maxillary lateral incisor was congenitally missing, the contralateral tooth was often peg-shaped. Grahnén (1956) further suggested, in his family study, that there was strong evidence to assume that a peg-shaped maxillary lateral incisor is a modified manifestation of hypodontia. Lai and Seow (1989) reported that peg-shaped maxillary laterals were observed in 8.9% of the hypodontia group, whereas there was no subject with this trait in the control group. Furthermore, the aforementioned authors reported a pattern in that 50% of those with peg shaped maxillary lateral incisors also had missing premolars only, whereas, in those with multiple missing teeth the incidence of maxillary peg-shaped lateral incisors was reported to fall to 16.7% (Lai and Seow 1989).

1.6.5.2 Altered root morphology.

1.6.5.2.1 Taurodontism.

There is an increased likelihood of taurodontism of the permanent mandibular first molars in the presence of hypodontia (Rune and Sarnas 1974, Schalk-van der Weide et al 1993a, Seow et Lai 1989, Arte et al 2001).

Taurodontism literally means "bull-like teeth" and it is accepted that taurodontism describes teeth which have an enlarged body of the tooth at the expense of the roots (Keith 1913, Larmour et al 2005). They characteristically show an enlarged and elongated pulp chamber (Larmour et al 2005).

An incidence of 6.3% in British schoolchildren with taurodontism has been reported (Holt and Brook 1979). From subject records, taurodontism of the mandibular first permanent molar was observed in 34.8 % of those with hypodontia compared to 7.5% in the controls (Seow and Lai 1989). Furthermore, the incidence of taurodontism increases with an increase in severity of hypodontia as seen in the Table 1.7 overleaf (Seow and Lai 1989).

Table 1.7 The incidence of taurodontism in relation to severity of hypodontia and hypodontia of specific tooth type (Seow and Lai 1989).

Type of hypodontia	Percentage of subjects showing taurodontism in mandibular first permanent molar (s)
Multiple missing teeth	56.5%
Missing premolars	30.4%
Missing lateral incisors	8.7%
Missing both laterals and premolars	4.3%

Schalk-van der Weide et al (1993a) reported that in ninety subjects with severe hypodontia, 28.9% showed taurodontism of one or two permanent mandibular first molars. The incidence of taurodontisn in the control group who had a full dentition was reported as 9.9%. Furthermore, of those with hypodontia, 10.8% of the mandibular first molars with taurodontism showed shortened roots.

1.6.6 Normal dental development.

Knowledge of the normal expected age range of dental development of all the permanent is required in order to correctly diagnose hypodontia with the aid of radiographs. Normal maturation of the development of the teeth will vary, due to individual, familial and racial differences. For example, as a general rule, females

mature dentally earlier than males and Africans more rapidly than Caucasians (Jorgenson 1980). Crown development of all permanent teeth is normally completed by age 10 years old (Garn et al 1959). Table 1.8 below shows the approximate ages at which teeth can be expected to be visible on radiographs.

Table 1.8 Age at which teeth are visible on radiographs (Jorgenson 1980).

Primary Teeth			Permanent Teeth		
	Maxillary	Mandibular		Maxillary	Mandibular
Central Incisor	Birth	Birth		6 months	6 months
Lateral Incisor	Birth	Birth		1 year	9 months
Canine	Birth	Birth		6 months	6 months
First Molar	Birth	Birth	First Premolar	2 years	2 years
Second Molar	Birth	Birth	Second Premolar	3 years	3 years
			First Molar	Birth	Birth
			Second Molar	4 years	4 years
			Third Molar	9 years	9 years

1.6.6.1 Delayed dental development and hypodontia.

Delay in normal dental development often occurs in those with hypodontia (Garn and Lewis 1961 and 1970, Gravely and Johnson 1971, Rune and Sarnas 1974, Schalk van der Weide et al 1993b).

Gravely and Johnson (1971) reported in their study of three pairs of twins that for all, there was delay in dental development. Garn and Lewis (1961) reported a delay in tooth formations of the remaining teeth, especially the most posterior teeth, in those with third molar hypodontia. Indeed, they reported on a posterior to anterior gradient of delay in tooth formation timing. However, Rune and Sarnas (1974) and Schalk van der Weide et al (1993b) in their studies were unable to ascertain this gradient.

In a controlled study that compared ninety-one children with advanced hypodontia to children without hypodontia, it was found that eighty-five of the hypodontia group showed delayed tooth formation in relation to chronological age. The range of the developmental timing differed from the controls in a range from 2.4 years ahead to 7.8 years delayed, with a mean of 1.8 years delay for boys and 2.0 for girls. Furthermore, despite considerable variation from one child to another being observed, the degree of delay was approximately the same for all the remaining teeth. It was further reported, that there was a tendency for the contralateral teeth to those missing to show marked delay in development (Rune and Sarnas 1974). Despite, Schalk-van der Weide et al (1993b), confirming a delay in tooth formation in those with hypodontia, they, however, found it occurred more often in males.

Furthermore, Aasheim and Ögaard (1993) reported that 11.3% of males who were diagnosed with hypodontia of second premolars at the age of 9 years old, had delayed enamel mineralization compared to 2.9% in girls. Therefore, it has been suggested that studies regarding prevalence of hypodontia should not be done on children younger than 9 years old because there has to be consideration given to the late development of the second premolars in boys (Magnusson 1977, Daugaard-Jenson et al 1997b, Niswander and Sujaku 1963, Wisth et al 1974).

Bailit et al (1968) reported no significant differences in the two populations they studied with regard to hypodontia and delayed dental development. However, the aforementioned authors do comment that there was sampling differences between their study and that of Garn and Lewis (1961) and this may account for the difference in findings of the two studies.

1.6.7 Abnormalities of the occlusion associated with hypodontia.

1.6.7.1 Malposition of canines.

Malpositions of canines and hypodontia occur together frequently.
Hypodontia has been found to be associated with palatally displaced canines (PDC), which are often found to be impacted (Peck et al 1994, 1996a, Pirinen et al 1996, Chaushu et al 2002), and transposed (Peck et al 1993, 1996b, 1998, Shapira and Kuftinec 2001). In Europeans, the palatally displaced canine has been found to occur in 1-3% of the population (Peck et al 2002) and two to three times more frequently than buccal displacement (Peck et al 1994). Transposition involves the positional interchange of two permanent teeth within the same quadrant of the

dental arch and are rare occurrences with the following the most frequently reported. The interchange of the maxillary canine with the first premolar (Mx.C.P1) having a prevalence of 0.25% (Peck et al 2002) and the interchange of the mandibular lateral incisor with the canine (Mn.L2.C) having a prevalence of 0.03% (Peck et al 2002).

Like hypodontia, the palatally displaced canine and transposition of the canine has been reported in families and has been suggested to be under genetic control (Peck et al 1993, 1994, 1998, Pirinen et al 1996, Svinhufvud et al 1988). However, other authors take the view that the genetically determined anomalies of the lateral incisor provide an environment that deprives the erupting canine of its normal guidance, labelled the guidance theory of palatal canine displacement (Brin et al 1986, Becker et al 1999). Becker (1999) suggested that the phenomenon of palatally displaced canines if genetically determined should occur at least equally or if not more on the side of the missing lateral incisor as it is of a greater expression of the gene abnormality. In contrast, buccal displacement of canines is usually due to inadequate space and in most cases the canines erupt (Peck et al 1994).

1.6.7.1.1 Palatally displaced canines.

In the presence of a PDC there is a significant increase in the frequency of hypodontia of molars and premolars but not the maxillary lateral incisor (Peck et al 1996 a and b).

There is general agreement that palatal displacement of maxillary canines occurs with greater frequency in subjects with peg-shaped or diminutive lateral incisors than in those with congenitally missing lateral incisors (Becker et al 1981, Becker 1999, Peck et al 1996a, 1996b, Chaushu et al 2002). Brin et al (1986) reported that if a subject had a small or peg shaped maxillary lateral incisor there was a one in ten probability that the canine would be palatally situated and a one in twenty probability if the maxillary lateral incisor was congenitally missing. Furthermore, Pirinen et al (1996) reported that PDC is related to incisor-premolar hypodontia and peg shaped incisors.

1.6.7.1.2 Transposition of canines.

Peck et al (1996b) reported that in Mx.C.P1, congenital absence of third molars accounted for 19%, which was approximately a normal rate, whereas absence of the maxillary lateral incisor accounted for 26%, showing a thirteen fold increase.

Peck et al (1998) reported that there was a statistically significant increase in the congenital absence of third molars and mandibular second premolars, and the presence of peg-shaped maxillary lateral incisors in association with Mn.L2.C

1.6.7.1.3 Orofacial fields.

Peck et al (1996b) suggested that the observed differences in the specific site of the hypodontia associated with PDC and Mx.C.P1 are associated with the suppression of tooth formation in odontogenic fields (specific sites of tooth development under a genetic influence). They suggested that the third molar

hypodontia associated with PDCs and Mn.L2.C represented the effect of a posterior orofacial field, and Mx.C.P1 with maxillary lateral incisor hypodontia represented the effect of an anterior orofacial field. Hypodontia of mandibular second premolars reported in PDCs, Mx.C.P1 and Mn.L2.C appear to represent an intermediate field (Peck et al 2002). These authors further suggested that the transcription factors, Msx 1 and Pax 9, which have been associated with hypodontia of molars might have a genetic involvement with PDC and Mn.L2.C with the specific expression of third molar hypodontia in the posterior orofacial field (Peck et al 2002). Pax 9 transcription factor has been associated with tooth bud positioning at the mesenchymal level and there may be a possibility that this genetic influence be involved in PDC and transpositions (Neubuser et al 1997, Peck et al 2002).

1.6.7.2 Reduced alveolar development.

Teeth are of paramount importance not only for the growth of alveolar bone but also for the maintenance of height of the alveolar bone. Where there is congenital absence of permanent teeth impaired alveolar bone development is frequently observed (Kurol and Thilander 1984a). The alveolar ridge appears thin and "wasted" as a result of the lack of teeth and the associated failure of alveolar bone. The sequelae in the posterior regions of those with severe hypodontia is lack of posterior support. This leads to an increase in the freeway space and overclosure of the dentition (Hobkirk et al 1994).

1.6.7.3 Infraocclusion of primary molars.

Infraocclusion of primary molars is strongly associated with hypodontia of the succedaneous premolar (Kurol and Thilander 1984a, Lai and Seow 1989, Antoniades et al 2002). Lai and Seow (1989) reported from their study of 1032 subject records that 65.7% of sixty-six subjects with hypodontia showed infraocclusion of primary molars compared to only 1.5% of the control children. Furthermore, in those subjects with multiple congenitally missing teeth infraocclusion accounted for 52.3% of the cases. Similarly, Antoniades et al (2002) reported that congenital absence of the successor tooth was observed in 54% of infraoccluded primary molars. The aforementioned authors further reported that, in infraocclusion of the first primary molar absence of the permanent successor was up to 30%, whilst this percentage rose to 63% in cases of second primary molar involvement.

It has been observed that mandibular first and second primary molars are most affected and it has been suggested that the occurrence of this infraocclusion varies between 8-14 % in the 6-11 age group (Kurol and Thilander 1984a). Antoniades et al (2002) have reported that infraocclusion affected the mandibular second primary molar more often than first primary molar.

1.6.7.4 Double primary teeth.

The most common effect of double teeth in the primary dentition is hypodontia of the associated permanent dentition (Munro 1958, Grahnén and Granath 1961, Nik-Hussein and Majid 1996). Niswander and Sujaku (1963) in his study of 4,150 Japanese children and Nik-Hussein and Majid (1996) observed hypodontia in the

permanent dentition of 58% and 50% respectively following the presence of double teeth in the primary dentition.

1.6.7.5 Rotated premolars.

Rotated premolars occur more frequently in those with hypodontia (Baccetti 1998b, Arte et al 2001). Baccetti (1998b) reported that this trait exceeded two to three times that of the normal population (3-5%).

1.6.7.6 Enamel hypoplasia.

There is a significant relationship between enamel hypoplasia and hypodontia (Lai and Seow 1989, Baccetti 1998a). Moreover, it was found that subjects with multiple congenitally missing teeth were responsible for 75% of all cases of enamel hypoplasia (Lai and Seow 1989).

1.6.8 Familial inheritance of hypodontia.

Chung et al (2000) reported that, of 59 subjects attending a Hypodontia Clinic, at Newcastle Dental School, United Kingdom, 37% confirmed a positive family history of hypodontia. Rose (1966) in his study of 6,000 subjects referred to an orthodontic practice, found that of the 258 (4.3%) presented with hypodontia, 10% gave a positive family history of congenitally missing teeth. In an earlier study, of 10,000 schoolchildren inspected by the school dental clinic, Dolder (1937) found that 5% of subjects with hypodontia gave a positive family history. The latter, is probably a more accurate account of the likelihood of a family

history in children presenting with hypodontia as this sample was more akin to the general population.

Grahnén (1956) showed that not only did the parents of individuals with hypodontia have a higher frequency themselves but their siblings also had a higher hypodontia frequency:- 26% compared to a population normal of 6%. Furthermore, he reported that the risk of hypodontia also increased in siblings as the number of missing teeth increased. In addition to this, Grahnén (1956) also reported that similar teeth and regions of the mouth were affected within families. Brook (1984) not only confirmed the findings of Grahnén (1956) but he also reported that the more severe the hypodontia, the more relatives were affected.

These family studies indicate that hypodontia is genetically determined (Grahnén 1956, Rose 1966, Dolder 1937). This genetic link was further highlighted in the study of 200 Finnish Skolt-Lapps, aged 8-16 years, where, not only was the prevalence of hypodontia in this genetically isolated population found to be 17.5%, 2-3 times higher than other mixed Scandinavian countries, but the malformations of the maxillary lateral incisor was also correspondingly high (Ingervall and Hedegaard 1975).

1.6.8.1 Accuracy of family history reporting.

Gaining an accurate family history is difficult. Errors are likely due to the reliance on accounts from family members (Graber 1978). Despite this, Saito (1959) and Jorgenson (1980) still found that relatives of those with hypodontia have a significantly higher rate of dental anomalies compared to the general population.

1.6.8.2 Association between hypodontia and other syndromes.

There are well known associations between hypodontia, in both the primary and permanent dentitions, and some syndromes (Lucas 2000). Indeed, hypodontia is an important diagnostic sign for these affected subjects (Graber 1978, Daugaard-Jenson et al 1997a). Some such as Böök's syndrome and Rieger's syndrome have specific patterns of missing teeth (see page 76, Table 1.10) whilst others have a generalised pattern, for example, Down syndrome, Hypohidrotic ectodermal dysplasia and Incontinentia pigmenti (Witkop et al 1975, Graber 1978, Schalk-van der Weide et al 1994, Daugaard-Jenson et al 1997a). Of the syndromes which feature hypodontia, the greatest number are of ectodermal origin, such as Hypohidrotic ectodermal dysplasia, Tooth and nail/Witkop syndrome, Incontinentia pigmenti, Bröök's syndrome, Rieger's syndrome, clefting syndromes and syndromes associated with growth and development defects such as Solitary Median Maxillary Central Incisor. Indeed, there are over one hundred syndromes that are characterised by dysplasia of ectodermal origin (Lucas 2000).

1.6.8.2.1 Ectodermal dysplasia.

Ectodermal dysplasia describes a group of syndromes which involve abnormal disturbances in structures and tissues derived from the ectodermal tissue layer, such as hair, teeth, skin and sweat glands (Ellis and Ahmed 1993, Kere et al 1996). Although rare, the most frequently reported and clinically distinct of the ectodermal dysplasia syndromes is that of X-linked recessive condition, anhidrotic (absence of sweat glands) or hypohidrotic (reduction in number of sweat glands) ectodermal dysplasia (Ellis and Ahmed 1993, Kere et al 1996). However, an

autosomal recessive (Ellis and Ahmed 1993) form of hypohydrotic ectodermal dysplasia condition has been reported.

1.6.8.2.1.1 Hypohidrotic ectodermal dysplasia.

An approximate prevalence for hypohidrotic ectodermal dysplasia is one in 10,000 births (Lucas 2000). These affected subjects show the characteristics of dry skin (hypohydrosis), hypodontia or anodontia, and sparse, fine hair (hypotrichosis) (Ellis and Ahmed 1993, Kere et al 1996). The hypodontia occurs in both dentitions and is generalised and severe, often involving in the permanent dentition, the maxillary central incisors and first molars, generally considered the most "stable teeth" – the least likely to be absent (Schalk-van der Weide et al 1994). Furthermore, any teeth that are present are often small (microdontia) and conical (misshapen) with hypoplastic/ hypomineralised enamel (Witkop et al 1975, Schalk-van der Weide et al 1994, Vierucci et al 1994, Jorgenson 1980). The alveolar ridges and maxillary tuberosities are poorly developed and subjects often have distinctive facial features, amongst which is a reduced lower face height (Ellis and Ahmed 1993, Vierucci et al 1994). The affected males usually show severe manifestations of the syndrome, whilst females demonstrate signs and symptoms varying from those as severely affected as males, to those only showing slight involvement, for example, a missing tooth or peg shaped tooth, or undetectable physical signs (Carter and Bordy 1967).

1.6.8.2.1.2 Tooth and nail syndrome or Witkop syndrome.

The simplest form of ectodermal dysplasia is tooth and nail syndrome, also known as Witkop syndrome, which is inherited as an autosomal dominant transmission. It is characterised by hypodontia and/or conical teeth and poorly formed and brittle nails. There is however, little involvement of the hair and of the sweat glands (Hudson and Witkop 1975). The aforementioned authors reported a pattern of missing mandibular incisors, second molars and maxillary canines in these subjects.

1.6.8.2.1.3 Incontinentia pigmenti.

Incontinentia pigmenti or Bloch-Sulzberger syndrome is rare and teeth anomalies occur in about 90% of subjects (Lucas 2000). Hypodontia and peg-shaped or conical teeth occur with both the primary and permanent dentitions affected (Welbury and Welbury 1999, Lucas 2000). Furthermore eye lesions occur in about 30% of subjects (Lucas 2000).

1.6.8.2.1.4 Bröök's syndrome.

The specific pattern of hypodontia of premolars in association with premature whitening of the hair and increased sweating are recognised features of Brööks syndrome (Bröök 1950).

1.6.8.2.1.5 Rieger's syndrome.

Rieger's syndrome includes ocular features such as goniodysgenesis (a developmental abnormality of the anterior ocular segment) with a specific pattern

of hypodontia of the maxillary incisors and failure of the periumbilical skin to involute (Jorgenson et al 1978). Owing to the specific pattern of hypodontia early recognition is possible before the associated glaucoma and its effects occur (Jorgenson 1980).

1.6.8.2.1.6 Solitary Median Central Incisor.

The presence of a solitary median central incisor (SMMCI) is rare and this condition has been reported as an isolated dental finding and also in association with holoprosencephaly (type of brain malformation), pituitary dysfunction and a large number of midline developmental defects (Cho and Drummond 2006).

1.6.8.2.2 Cleft lip and cleft palate.

Cleft lip and cleft palate are fusion disorders that affect the midfacial skeleton and can either occur in isolation or as a feature of some syndromes. Hypodontia is a feature of patients with cleft lip and/or palate, with the maxillary lateral incisor being reported the most frequently missing tooth excluding the third molars in the cleft and the second premolars outside the cleft. The likelihood of hypodontia of the maxillary lateral incisor increases with the severity of the cleft (Shapira et al 2000). Lekkas et al (2000) reported that their findings supported the hypothesis that surgery for closure of the hard palate in early childhood was a factor responsible for hypodontia outside the cleft area, due to disruption of the developing tooth buds.

1.6.8.2.3 Down syndrome.

Down syndrome or trisomy 21 is a common chromosomal disorder and the overall prevalence rate is approximately 1:800 live births. The risk increases with maternal age, reaching 1:50 for mothers over 45 years of age (Lucas 2000). Hypodontia is a common feature of subjects with Down syndrome with the maxillary lateral incisor reported as the most commonly missing (Meštrović et al 1998, Lucas 2000, Acerbi et al 2001). Teeth that are present are often hypoplastic and conically shaped Lucas 2000).

1.7 Aetiology of hypodontia.

1.7.1 Introduction.

The most likely factors to cause hypodontia are of evolution and inheritance (Graber 1978). However, trauma, infection of the developing tooth bud, radiation, glandular dysfunction, systemic conditions such as rickets or syphilis, German measles during pregnancy and severe intrauterine disturbances have all been proposed as possible causal factors in hypodontia (Graber 1978, Burzynski and Escobar 1983, Nunn et al 2003).

1.7.2 Normal tooth development.

In all mammals the teeth form specific groups defined by their shape, size and position in the dental arch, for example, incisors, canines, premolars and molars (Cobourne 1999). The early stages of tooth development are similar irrespective of the type of tooth being formed (Peters and Balling 1999). The formation of the

different shapes of teeth is called morphogenesis and their correct positioning in the jaw is known as patterning (Cobourne 1999).

The formation of a tooth is the result of a complicated growth process, which begins by the 6th week of intra-uterine life, and involves proliferation and differentiation of cells. The proliferation of cells from the epithelial lining of the oral cavity (oral ectoderm) form outbuddings, referred to as the dental lamina, which undergoes various stages of development, forming a tooth germ, from which eventually develops a tooth (Langman 1974). These outbuddings, of the dental lamina into the underlying mesenchyme form epithelial buds, known as the bud stage. During this process the mesenchymal cells gather around the bud (condense) and form the dental papilla which eventually results in the formation of the tooth pulp and the dentine secreting odontoblasts. Migration of mesenchymal cells around the epithelial bud forms the dental follicle. The epithelial component undergoes specific stages during the cap and bell stages and eventually gives rise to ameloblasts, which form the enamel (Peters and Balling 1999). The stages for tooth development are the same for both primary and permanent teeth, with the latter developing lingual to the former. However, the first, second and third molars are formed from extensions of the dental lamina (Langman 1974).

During development, much of the mesenchyme of the orofacial region is provided by migration of neural crest cells from the hindbrain and it is, to these cells that researchers in the field of molecular biology are investigating in regards to their possible pre-specification or as a controlling mechanism controlling this pre-

specification (Cobourne 1999). This is of special interest, as the maxilla and mandible develop differently: the mandibular dentition develops in a continuous dental lamina in the mandible (mandibular branch of the first branchial arch), whereas the maxillary incisors develop in the medial nasal processes, and the other maxillary teeth develop in two separated maxillary branches of the first arch. Subsequently, the maxillary branches and the medial nasal processes fuse to form the palate and the maxillas. Therefore, it seems that any controlling mechanism must occur at a very early stage in development to account for the regionalisation (Weiss et al 1998).

1.7.3 Dental Evolution.

Research in dental anthropology provides insight into the evolutionary aspects of tooth development and has given rise to theories relating to hypodontia.

Initially teeth were identical conical individual structures called "homodonty", from which "heterodonty" evolved in mammals resulting in a diverse dentition (Butler 1995). Teeth are vertebrate-specific and within vertebrates, species-specific. Tooth shape, varies with position in the jaws and is bilateral and symmetric (Vastardis 2000). Teeth and teeth bearing bones evolve together (Butler 1995). Indeed, the reduction in the size of the jaws together with a reduction in tooth number is believed to be a continuing evolutionary trend (Dahlberg 1945, Vastardis 2000). Lavelle et al (1970) reported a tendency, in humans, towards a shortened maxillomandibular skeleton compared to their predecessors.

1.7.3.1 Evolutionary and anatomical theories.

Evolutionary and anatomical models are attempts to explain the occurrence of hypodontia. Butler (1939), proposed that the mammalian dentition can be divided into three developmental fields; molars/premolars, incisors, and canines. Within each field there is a "key" tooth, which is more stable developmentally. The remaining teeth on either side of this tooth become progressively more unstable, the more distant their position from the "key" tooth in that field, resulting in greater variation in size and shape, and an increased likelihood of being "evolutionary lost" (Butler 1939).

Thus, in the human dentition, according to Dalberg (1945), the "key" tooth in the fields; incisor, canine, premolar and molar are the mandibular lateral incisor, maxillary central incisor, the canine, the first premolar and first molar. This was based on the findings that the maxillary lateral incisors, the second premolars and the third molars were more likely to be missing and more vulnerable to modifications in shape and size. The "key" tooth was defined as the most mesial in each field, except for the mandibular lateral incisor, which is reported as being more stable than the central incisor. Furthermore, Bailit (1975) reported that not all teeth are equally heritable and that the "key" tooth has the highest heritability and is the more stable tooth morphologically. Indeed, Schalk-van der Weide et al (1992) concur with this earlier literature and reported in their study that, the more distal teeth seemed to be more affected in terms of morphology, size, eruption and number.

Bailit (1975) further reported that the distal tooth in the field seems to be influenced more by the environment. In fact, according to the aforementioned author, the severe reduction in size of the lateral incisors and second premolars should be mostly due to environmental factors. However, it is difficult to explain why teeth developing at different times are affected by the same environmental factors, which suggests a genetic influence being more likely (Schalk-van der Weide et al 1992).

Other authors, however, feel that the result of the more severely reduced distal teeth is more likely to be an interaction of genetic and environmental factors – referred to as polygenic mode of inheritance (Suarez and Spence 1974, Brook 1984).

1.7.4 The genetic aetiology.

Hypodontia can occur in association with genetic syndromes, or an isolated sporadic or familial trait (Graber 1978, Gorlin et al 2001, Jorgenson 1980). The literature supports that inheritance plays a role in hypodontia. However, the mode of inheritance is still a controversial issue (Burzynski and Escobar 1983). Grahnén (1956) reported that in the majority of cases of familial hypodontia the mode is mostly likely to involve a single dominant gene. Alvesalo and Portin (1969) reported that missing and peg shaped maxillary lateral incisors show an inheritance pattern of a single autosomal dominant gene with incomplete inheritance. Both, Grahnén (1956) and Alvesalo and Portin (1969) concur a peg shaped tooth is a weaker expression of the dominant gene. Furthermore, Alvesalo and Portin (1969) suggested that the degree of expression is determined by

modifying factors. Arte et al (2001) reported an autosomal –dominant transmission with reduced penetrance with respect to incisor-premolar hypodontia. Dahlberg (1937) reported hypodontia as an X-linked disorder, whereas, Thomsen (1952) reported a recessive mode of transmission, but this occurred in an isolated and inbred community. However, Alvesalo and Portin (1969), suggested that these modifying factors may lead to a mode of transmission resembling a recessive gene.

Affected members within a family can show variability in respect of, number of teeth involved, site, symmetry and the size, shape and rate of development of the remaining teeth (Graber 1978, Burzynski and Escobar 1983, Alvesalo and Portin 1969).

Nonetheless, Vastardis (2000) suggested that the interfamilial clinical variation of autosomal dominant traits may indicate that a multiplicity of gene defects may cause familial hypodontia. The aforementioned author, also hypothesised that this variation in clinical expression of familial hypodontia reflects genetic variability in the population.

In conclusion, it is, generally accepted that the mode of inheritance for familial hypodontia is that of an autosomal dominant pattern with incomplete penetrance and variable expressivity (Graber 1978).

Since sporadic or isolated hypodontia arise from a first mutation in a responsible gene, these cases lack a positive family history (Mostowska et al 2003a). The

aforementioned authors suggested that mutations in the gene Pax 9 could be responsible for this form hypodontia.

1.7.4.1 The genetic influences upon tooth development.

Tooth development is a very complex process and appears to involve many genes (Peters and Balling 1999, Vastardis 2000). These genes encode transcription factors and signal molecules which play an important role in the formation of many organs including teeth (Thesleff 1998, Peters and Balling 1999).

The understanding of the genetic control of tooth development at the molecular level has occurred as a result of investigations on the drosophila fly and mice (Cobourne 1999, Jowett et al 1993, Peters and Balling 1999). Investigations with mice, have indicated, that tooth development is regulated by interactions between epithelial and mesenchymal cells and is dependent on a number of genes (Thesleff 1998, Peters and Balling 1999). Furthermore, these investigations have given an insight into the signalling mechanisms that occur during tooth formation (Cobourne 1999).

The homeobox genes are a large group of genes which were originally discovered in the drosophila fruit fly and were found to contain a nucleotide sequence called the homeobox. One such gene isolated is the muscle segment homeobox (msh) gene (Mackenzie et al 1991). The murine equivalent of the msh-like genes are Msx 1 and Msx 2, which were formerly known as Hox-7 and Hox-8 respectively (Jowett et al 1993). Indeed, investigations have found that those mice deficient in the Msx 1 gene showed an arrest in tooth development at the bud stage, whilst

those deficient in Msx 2 gene, showed late defects in tooth development (Maas and Bei 1997). Jowett et al (1993) reported that Msx 1 is expressed exclusively in mesenchyme, both in the dental papilla and follicle, whereas, Msx 2 is expressed in the dental epithelium and only in the mesenchyme of the dental papilla. Indeed, the expression of both Msx 1 and Msx 2 in the dental mesenchyme requires the presence of the epithelium until the early bell stage and, furthermore, that the maintenance of the homeobox gene expression is dependent on epithelial and mesenchymal interactions.

It has been further reported that early expression of the Pax 9 gene not only marks the future site of tooth development, but preliminary analyses show that, Pax 9 is essential for the formation of teeth and is required for tooth development to proceed beyond the bud stage (Peters et al 1998a, 1998b). Indeed, Peters and Balling (1999) reported that the mesenchymal transcription factors Msx 1 and Pax 9 are initially regulated by epithelial fibroblast growth factors (FGFs) and bone morphogenic proteins (BMPs) and proposed a molecular model by which the interactions were controlled to explain their findings and also why teeth develop only at certain sites.

As previously discussed, the function of Msx 1 and Pax 9 is essential for tooth development to proceed beyond the bud stage. Peters and Balling (1999) reported that of all the FGF genes known, seven are expressed in the developing tooth, namely FGF 1,-2,-3,-4,-7,-8,-9. FGF 8 and FGF 9 expression, is detectable in the oral epithelium and both, are able to induce expression of Msx 1. FGF 8 can also in vitro be substituted for the inducing activity of the epithelium on Pax 9. FGF 8

is present in the oral epithelium and not restricted to the future sites of the developing tooth in the mouse. However, during initiation of tooth development Pax 9 is only expressed at those sites, therefore, a mechanism must exist to inhibit the expression of Pax 9 in those regions of the mandible where teeth will not develop. BMP 4 and BMP 2 (bone morphogenic proteins), both molecules of the transforming growth factor β (TGFβ) superfamily are able to antagonise Pax 9-inducing activity of FGF 8 in the tooth mesenchyme.

Based on these findings, Peters and Balling (1999), proposed that at the onset of tooth development Pax 9 expression is induced only at those sites at which FGF 8 / FGF 9 are expressed in the overlying epithelium and in which BMP 4 / 2 signalling does not interfere with Pax 9 inducing activities of FGF 8. In contrast, BMP 4 also stimulates the expression of Msx 1 and BMP 4 itself. Nothing is known about the regulation of BMP 4 at the initiation stage, however at the bud stage a little understanding is emerging. At this stage, Pax 9 and Msx 1 are co-expressed in the mesenchyme in which the function of both genes is required for the expression of BMP 4. The mesenchymal expressions of BMP 4 and Msx 1 at the bud stage act as a positive feedback loop. Most likely, a key function of Pax 9 and Msx 1 is the maintenance of the mesenchymal BMP 4 expression because mesenchymal BMP 4 signalling is involved in the induction of the enamel knot, a transient signalling centre of the epithelium that directs the next phase of tooth development (Peters and Balling 1999).

1.7.4.2 The genes.

Through genetic linkage in families, advancement in genetics and the human genome project, defective genes can be identified, allowing the mapping of inherited conditions (Vastardis 2000). Mutation of several genes associated with hypodontia in syndromes, including that of ectodermal dysplasias have been identified (Kere et al 1996, Semina et al 1996, Jumlongras et al 2001).

Ectodermal dysplasia is usually transmitted as an x-linked recessive trait, although it can be occur as an autosomal recessive and autosomal dominant trait. As a result of the x-linked recessive trait, a mother has a 50% chance of transmitting the affected gene to her son and a 50% chance of her daughter becoming a carrier. Female carriers of this gene will demonstrate minimal expression in the form of simple hypodontia and/or reduction in size of the dentition or only some teeth. Therefore, the dental profession is in a unique situation to be able to identify these subjects and has a responsibility to detect those who may be of carrier status to a potentially life threatening condition (McLaughlin 1991, Nunn et al 2003).

The Msx 1 gene is located on chromosome 4p16, whilst the Pax 9 is situated on chromosome 14 q21-q13 and belongs to the PAX gene family, of which there are nine (Peters et al 1998a, Mostowska et al 2003b).

Many candidate transcription factor genes, including members of the Pax and Msx, and other homeobox gene families, and a number of facilating factors such as Lef1 and Sox genes are expressed in teeth in different ways and probably are the downstream activated products of earlier signalling processes (Weiss et al

1998) . Weiss et al (1998) suggested that signalling factors like Sonic Hedgehog (SHH) and Pax transcription factors may be involved in patterning of the incisor region. Peters and Balling (1999) reported that SHH is released by the dental epithelium and induces the expression of the transcription factor Gli1 and the SHH receptor Patched (Ptc) in the mesenchyme. However in the absence of Msx 1, SHH fails to induce Ptc in the dental mesenchyme suggesting that during early tooth development the signalling of BMPs and FGFs which induce the expression of Msx 1 are required at least for the subset of the functional activities of SHH.

Sharpe (1995) postulated that a homeobox code may exist which may pattern the dentition. Furthermore, Thomas and Sharpe (1998) reported that, the overlapping expression of the genes Bar X1, DLx-2, Msx 1 and Msx 2 constituted an odontogenic homeobox code which patterns the dentition.

However, Weiss et al (1998) also suggested that rather than a genetic expression of a code, patterning may be the result of parameters of interaction among the genes. For example, tooth location may reflect a particular concentration of signalling factors rather than a set genetic programme. These may involve concentration-dependent or threshold response mechanisms.

1.7.5 The multifactorial influences in the aetiology of hypodontia.

There is literature to support the hypothesis that hypodontia is a polygenic multifactorial mode of inheritance, that is to say, the result of the interaction of many genes with environmental influences (Davis 1968, Suarez and Spence 1974, Brook 1984, Schalk-van der Weide et al 1992, Peck et al 1993, 1994).

The effects of genetic and environmental influences have been separated by comparing monozygous twins (identical). Differences between monozygous twin pairs should reflect environmental factors, whereas differences between dizygous twin pairs reflect both genetic and environmental factors (Lamour et al 2005). In general, a variable expression of hypodontia in monozygous twins was reported, suggesting an environmental influence (Gravely and Johnson 1971).

As previously mentioned, some studies proposed that hypodontia is caused by an autosomal dominant pattern (Grahnén 1956). However, clinically it is known that there is variable expression of the trait, suggesting there is a variable gene penetration, which is indicative of a polygenic mode of inheritance (Larmour et al 2005).

Identification of mutated genes that cause familial hypodontia will enable studies to assess the mechanism by which environmental factors modify gene expression and result in similar clinical phenotypes (Vastardis 2000).

1.8 Genes and the patterns of missing teeth.

A pattern of missing teeth can be described as a repeatable and type-specific sequence of congenitally missing teeth. Different patterns of missing teeth are caused by different mutated genes (Nieminen et al 1995, Vastardis et al 1996, Arte et al 1996, Stockton et al 2000).

In humans, it has been demonstrated that Msx 1 and Pax 9 mutations are associated with selective tooth agenesis (Vastardis et al 1996, Van Den Boogaard

et al 2000, Stockton et al 2000, Lideral and Reising 2002). Vastardis et al (1996) reported that, the mutation of Arginine to Proline substitution within the Msx 1 gene, resulted in hypodontia of the second premolars and third molars. Despite this, the effect of the Msx 1 mutation appears to be incomplete in other locations (Vastardis et al 1996). It has been suggested that the Msx 1 is not critical, or other transcription factors have compensated for this defect at these tooth positions (Nieminen et al 1995). Vastardis et al (1996) also suspected that, there is a redundancy of these transcription factors and/or other genetic mechanisms in the tooth formation and patterning of the primary dentition.

In general, different mutations in Pax 9 have been associated with absence of permanent molars (Stockton et al 2000, Nieminen et al 2001). Stockton et al (2000) reported in their study that, some individuals had hypodontia of most of the permanent molars despite having had a normal primary dentition. Furthermore, in addition to the lack of permanent molars, other individuals also lacked maxillary and/or mandibular second premolars as well as mandibular central incisors.

Mostowska et al (2003a) reported that their study confirms the hypothesis that more than one gene may be responsible for a specific pattern of hypodontia, as various mutations in Pax 9 give rise to the same phenotype. Furthermore, the aforementioned authors reported hypodontia in subjects with no mutation in either Msx 1 and Pax 9, which they suggested was a result of other genes encoding transcription factors which might affect the tooth patterning.

Msx 1 and Msx 2 have been shown not to be linked to the more commonly incisor –premolar hypodontia (Nieminen et al 1995). Futhermore, Arte et al (1996) reported that, some growth factors, namely fibroblast growth factor (FGF-3), epidermal growth factor (EGF) and the latter's receptor (EGFR), are not responsible for incisor-premolar hypodontia despite their implication of important functions in murine tooth development.

Vieira et al (2004) not only reported that there was evidence of an interaction between Msx 1 and Pax 9 but that there was a "borderline" suggestion that transforming growth factor alpha (TGFA) might play a role in cases where hypodontia included incisors. Vieira (2006) reported that, TGFA is a well characterised mammalian growth factor and that it has been mapped to chromosome 2p13. This is the only literature which has reported on the possible role of TGFA in hypodontia, especially that involving incisors.

A mutation identified in Msx 1 has been associated with hypodontia of premolars and the presence of orofacial clefts (Van Den Boogaard et al 2000). Furthermore, it has been reported that a mutation of Msx 1 is responsible for Witkop syndrome, which includes hypodontia and nail dysgenesis (Jumlongras et al 2001). However, in some syndromes, a specific pattern of congenitally missing teeth occurs (see Table 1.10, page 76) (Schalk-van der Weide et al 1994). Congenitally missing teeth have been described as either a significant diagnostic feature or an associated finding in more than 120 syndromes (Schalk-van der Weide et al 1994). The importance of this is essential especially in those cases where serious medical effects of the syndrome can be identified and/or alleviated or prevented.

Furthermore, The On-line Mendelian Inheritance in Man http;//www.ncbi.nlm.nih.gov/OMIM) has more than sixty syndromes listed associated with tooth anomalies (Vastardis 2000). However, there are other syndromes which are associated with hypodontia of a more generalised nature, for example Ectodermal dysplasia (Daugaard-Jenson et al 1997a). For these subjects the pattern of hypodontia may mimic non-syndromic hypodontia unless genetic analysis is performed.

So, to date there appears to be some genes that might link to the most commonly reported congenitally missing teeth and to the pattern of congenitally missing teeth. These might be summarised as follows in Table 1.9 overleaf.

Table 1.9 Summary of the possible gene markers to the pattern of congenitally missing permanent teeth.

Category	Pattern of permanent missing teeth	Possible gene responsible
1	Maxillary lateral incisors only	Unknown gene/perhaps TGFA
2	Second premolars and third molars	Msx 1
3	Molars alone	Pax 9-referred to as Pax 9m
4	Molars, second premolars and mandibular central incisors	Pax 9-referred to as Pax 9pi
5	Molars and incisors	Unknown/possible TGFA involvement
6	Combination of incisors	Unknown gene/perhaps TGFA
7	Canines alone Canines and third molars Canines, third molars and incisors	Unknown Unknown Unknown/possible TGFA involvement
8	Premolars only	Unknown
9	Premolars and incisors	Unknown/possible TGFA involvement
10	Others with severe hypodontia of molars and premolars with other teeth.	Unknown (possible undiagnosed syndromes)

At present the literature reports on prevalence, severity and the most common missing teeth rather than the pattern of missing teeth. To date there is no literature which has investigated the patterns of teeth in a sample of subjects presenting with non-syndromic hypodontia and attempted to match this to known genetic markers for hypodontia according to the literature.

Table 1.10 Syndromes with specific patterns of congenitally missing teeth (Schalk-van der Weide et al 1994).

Syndromes	Pattern of congenitally missing teeth	Mode of inheritance
Cleft lip / palate	Maxillary lateral incisors and canines	Multifactorial
Down syndrome	Third molars, mandibular second premolars and maxillary lateral incisors	Chromosomal
Rieger syndrome	Maxillary incisors (constant), and mandibular incisors and premolars (occasional)	Autosomal Dominant
Ellis van Creveld syndrome	Mandibular incisors and canines	Autosomal Recessive
Bröök's syndrome	Premolars	Autosomal Dominant
Gorlin-Chaudry-Moss syndrome	Primary molars, permanent premolars and molars	Autosomal Recessive
Lipoid proteinosis	Maxillary lateral incisors, canines and premolars	Autosomal recessive
Orofaciodigital syndrome Type 1	Mandibular incisors and canines	X-linked Dominant
Coffin-Lowry syndrome	Maxillary lateral incisors, mandibular incisors	X-linked Recessive
Holoprosencephaly	Maxillary incisors	Different modes
Hypoglossia-hypodactylia syndrome	Mandibular incisors and canines	Sporadic
Glossopalatine ankylosis syndrome	Incisors	Autosomal Dominant

1.9 Effects of hypodontia.

The effects of hypodontia have a profound implication for the management of subjects with this trait.

1.9.1 Function.

It has been reported that occasionally there was a reduction in masticatory function (Hobkirk and Brook 1980). However, subjects' complaints were towards a loss of aesthetics not function (Hobkirk et al 1994).

1.9.2 Aesthetic considerations.

The most obvious effect of congenitally missing teeth is that of spacing in the dentition with or without retention of primary teeth beyond their normal exfoliation time. The spacing and the lengths of the spans will increase with the severity of the hypodontia. In the anterior region of the mouth, the spacing and the mismatch of size of permanent teeth and primary teeth especially, if the latter are worn, may result in an aesthetically displeasing appearance (Hobkirk and Brook 1980).

Owing to the effects of the alteration in morphology of the dentition resulting in peg or conical shaped teeth dissatisfaction with appearance often occurs (Jepson et al 2003).

Subjects with hypodontia are often associated with a deep overbite, decreased interocclusal space and a reduced face height (Gravely and Johnson 1971,

Hobkirk and Brook 1980, Jepson et al 2003). Hypodontia can lead to midline asymmetry which has been reported as being less acceptable to the human eye (Millar and Taylor 1995).

It has been reported that dental caries may be less prevalent because of the spaced dentitions. However, in subjects with ectodermal dysplasia there is an increased risk of developing caries because of the associated xerostomia. Furthermore, the presence of enamel hyoplasia and hypomimeralisation is associated with an increase risk of caries incidence (Nunn et al 2003).

1.9.3 Psychological impact.

There is little research to evaluate the impact of extensive hypodontia on a young person (Hummel and Guddack 1997, Nunn et al 2003). It is reported that hypodontia can be distressing for subjects, with the missing and malformed teeth together with the facial appearance causing psychological upset (Hobkirk and Brook 1980) but the extent has not been evaluated. Peer group pressure may be factor with these subjects (Jepson et al 2003).

1.9.4 Treatment.

The lengths of the space/gap spans will impact on the treatment options (Hobkirk and Brook 1980). Space loss will occur if retained primary teeth become infraoccluded resulting in tipping of the adjacent teeth, especially the first permanent molars (Kurol and Thilander 1984a and b, Goodman et al 1994). The

ankylosis associated with infraocclusion has surgical and orthodontic implications (Goodman et al 1994).

The developmental delay of subjects with hypodontia, impacts on the timing of active treatment. As a result, restorative treatment is often delayed waiting for the eruption of teeth and root formation to occur (Jepson et al 2003). Furthermore, the teeth may erupt into undesirable positions (Jepson et al 2003, Schalk-der Weide et al 1993b). Rotated teeth are often in an unfavourable position for restorative treatment (Nunn et al 2003).

Overeruption of unopposed permanent teeth impacts not only on the vertical space available for the planned restoration but also on its success. Excursive movements of the occlusion when overerupted teeth are present can result in mechanical overloading and ultimately failure of the restoration (Jepson et al 2003).

Peg and conical shaped teeth because of the minimal crown height and small surface area, these teeth are not ideal for bridgework (Jepson et al 2003). Furthermore, the lack of undercuts with these microdont teeth present problems with retention with removable restorations (Goodman et al 1994, Hobkirk et al 1995). The manifestations of the presence of taurodontism are that endodontic therapy can be difficult due to the shape of the pulp chamber and location of the pulp canals. The unfavourable crown-root ratio increases the likelihood of bridgework and prosthetic treatment to fail (Schalk-van der Weide et al 1993a).

The limitation on the quality and quantity of alveolar bone has major implications for restoration of the space with osseointegrated implants. There may be insufficient bone to provide an implant as doing so would increase the risk to such structures as the inferior dental nerve and the maxillary sinus. Furthermore, a high crown : implant ratio would be unfavourable and also produce poor aesthetics (Jepson et al 2003).

Late referral often occurs at the age of adolescence and results in treatment coinciding with the important years for examinations. Time off from school and studies is often difficult at this time (Hobkirk et al 1994). Children with hypodontia may suffer from social isolation and may also have poor co-operation for dental treatment (Nussbaum and Carrel 1976, Nunn et al 2003).

1.10 Management of hypodontia.

The aim of management and treatment of the subject with hypodontia is no different than that of a non-hypodontia subject. These aims are the restoration, maintenance or improvement of function and appearance (Jepson et al 2003). However, as a result of the profound effects of hypodontia, the management and treatment is often complex and involves different specialist skills which not one operator or department may possess. Therefore, a multidisciplinary team approach involving restorative (Paediatric dentistry), orthodontic and maxillofacial input is desirable for an optimum outcome of treatment (Hobkirk et al 1994, Bergendal et al 1996, Nunn et al 2003, Jepson et al 2003). The management of the subject with hypodontia can be challenging as their management often requires long-term treatment which reflects their age, needs and limitations (Jepson et al 2003).

Careful clinical assessments by the team with often the additional use of diagnostic wax ups are required so as, to provide the desired aesthetic and occlusal treatment plan (Jepson et al 2003). The treatment is dependent on a number of factors namely the pattern of missing teeth, the residual spacing, the malocclusion and of course the subject's attitude.

In general terms treatment falls into three broad categories as follows:

- Accept spacing
- Space closure
- Redistribution of space

(Jepson et al 2003).

1.10.1 Behavioural management.

The success of treatment depends not only with the expertise of the team members and the quality of materials available for use but with the subject and their parents/guardians. The team can aid both the subject and parent to achieve a successful outcome but factors such as the subject's ability to accept and implement advice, change undesirable habits and accept treatment bring its limitations (Jepson et al 2003). Treatment can often involve many appointments and time for these has to be anticipated and accommodated in a busy life with work, exams, and social commitments (Hobkirk et al 1995, Jepson et al 2003). Furthermore, subjects must have realistic expectations of what can be achieved and be aware of the limitations of the treatment (Millar and Taylor 1995).

1.10.2 Maintenance of the existing dentition.

1.10.2.1 Preventive support.

As with any subject dietry advice, oral hygiene instruction of the dentition and any appliances, preventive measures (Fluoride toothpaste, Fluoride rinse, Fluoride tablets, fissure sealents, topical Fluoride applications) and regular review is paramount (Nunn et el 2003).

1.10.2.2 Management of the retained primary dentition.

Where the succedaneous permanent tooth is missing, it is beneficial to retain the existing primary dentition to retain bone for either transplantation or implants. Primary teeth also act as the ideal space maintainers, preventing undesirable drifting of adjacent teeth which may result in problems with definitive treatment at a later date (Hobson et al 2003). Therefore, the appropriate treatment for caries and pulpal pathology should be undertaken (Nunn et al 2003). Space maintenance can be provided with the use of stainless steel crowns or band bonded with a wire loop (Hobson et al 2003). Often however, according to the literature, these primary teeth can be diminutive or infraoccluded. As a result, their management may require "intermediate" restorations with composite additions on the diminutive teeth and onlays (ceramic, composite or gold) on the infraoccluded teeth (Cavanaugh and Croll 1994, Evans and Briggs 1996, Nunn et al 2003). Extraction of these infraoccluded primary teeth is indicated if it is situated below the interproximal points of the adjacent teeth and also before it becomes ankylosed, as extraction at this point would result in excessive bone loss (Hobson et al 2003). Retention of primary dentition is unpredictable (Haselden et al 2001)

and their eventual loss should be anticipated and built into the treatment plan (Jepson et al 2003).

1.10.3 No corrective treatment.

The residual spacing may be minimal and/or may occur posteriorly and as a result is not of any aesthetic concern to the subject, resulting in the decision of no corrective treatment. Indeed, the subject may be unwilling to have the necessary corrective treatment (Jepson et al 2003).

1.10.4 Orthodontic treatment.

Space closure can also be achieved through orthodontic treatment only or can be combined with composite build up and veneers, bridgework or implants. Orthodontic treatment may be required to position the remaining teeth, especially those cited as abutment(s) to favourable positions (Hobkirk et al 1995, Jepson et al 2003).

1.10.5 Restorative management.

1.10.5.1 Bonded composite build-ups and veneers.

The appearance of malformed primary or permanent anterior teeth together with the spacing associated with hypodontia can be improved using bonded composite build-ups and veneers (Jepson et al 2003, Lambert 2006). Although porcelain is aesthetically superior to composite as a veneer, apical migration of the gingival

margin in young patients rapidly results in a poor aesthetic appearance (Welbury et al 2005).

These composite restorations can be considered minimally invasive and as such are a popular conservative treatment option for adolescents (Lambert 2006) and can be achieved by either a direct or an indirect technique (Evans and Briggs 1996, Lambert 2006). This form of treatment is often used to camouflage canines to resemble maxillary lateral incisors. Success of this is dependent on the colour and being able to modifying the shape of the canine with reduction. Another consideration is the more apical gingival margin of the canine compared to the maxillary central incisors. As a result in subjects with high smile lines the aesthetics may be compromised. Furthermore, canines positioned in the maxillary lateral incisor position alter the canine guidance in lateral excursions (Millar and Taylor 1995, Jepson et al 2003).

Furthermore, Lambert 2006 reported on the use of a fibre-reinforced direct composite bridge to treat anterior spacing as a result of hypodontia. Indeed, the author further reported that this composite bridge could either function as a "transitional" restoration for those awaiting an implant at the appropriate age or as a definitive treatment.

1.10.5.2 Resin-retained bridgework.

Adhesive rather than conventional bridgework is preferred in unrestored teeth especially those of immature teeth with large pulps (Hobkirk et al 1995, Jepson et al 2003). The longevity of these teeth is paramount in these subjects.

Unfortunately, the retention of bridgework is compromised in the presence of short clinical crowns especially that of retained primary teeth. Another major factor in the provision of bridgework is the available vertical space. The preferred option is a cantilever design supported by a single abutment and pontic. In cases involving orthodontic treatment full time retention of six months would have to be completed prior to construction of a resin-retained bridge. A consideration is also that repair or replacement of this type of bridge is relatively simple (Jepson et al 2003).

1.10.5.3 Conventional bridgework.

This is contraindicated in young subjects because of the tooth reduction required and the risk of pulpal exposure. However, in older subjects it is acceptable especially if the existing dentition is restored (Jepson et al 2003).

1.10.6 Prosthodontics.

There is evidence that longterm use of dentures causing harm to the remaining teeth (Carlsson et al 1965, Jepson et al 2003), although with simple designs and good oral hygiene this can be reduced (Millar and Taylor 1995). In the absence of good oral hygiene and diet, problems of caries and periodontal disease can arise with the use of overdentures (Hobkirk and Brook 1980). Use of topical fluoride application within overdentures has been reported to reduce this caries risk (Renner and Kleinerman 1980). However, dentures have a value as an interim treatment prior to a definitive treatment. In this situation, partial acrylic dentures are more often used than cobalt chrome partial dentures to replace missing teeth

and improve aesthetics. Additionally, they can be used as space maintainers after orthodontic treatment or to prevent overeruption of unopposed teeth. Overdentures allow improvement in function but also in aesthetics, by not only replacing missing teeth but by restoration of the face height. Furthermore, dentures can accommodate further growth and eruption of teeth with easy adjustments. The pros and cons of provision of a denture are dependent on the subject's individual needs and willingness to wear it (Jepson et al 2003).

1.10.7 Oral Surgery.

1.10.7.1 Primary extractions.

Carefully timed extractions of primary teeth can achieve spontaneous space closure (Millar and Taylor 1995, Mamopoulou et al 1996). However, prior to any primary extractions, consideration of an existing malocclusion must be taken into account (Millar and Taylor 1995).

1.10.7.2 Surgical Exposure.

Unerupted ectopic teeth in the subject with hypodontia can be surgically exposed and where required a variety of orthodontic attachments can be placed to bring the tooth into aesthetic alignment in the arch (Bishara 1992, Meechan et al 2003)

1.10.7.3 Implants.

Implants are the preferred definitive choice of treatment for subjects with hypodontia, particularly where the dentition is spaced and microdont. However,

the outcome in respect of implant and tooth position may be compromised in the subject with hypodontia because of the limitations of the alveolar bone (Jepson et al 2003, Sweeney et al 2005). Indeed, bone augmentation may also be required in addition to the provision of the implant to reduce these limitations of the alveolar ridge (Jepson et al 2003, Meechan et al 2003). Bone augumentation can be achieved by bone grafting from another area of the subject, usually the iliac crest of the hip or in combination with alloplastic materials (Meechan et al 2003). These are normally placed after the period of growth has completed, so as to avoid inhibition of ridge formation in these developing ridges and ankylosis of the implant (Meechan et al 2003, Sweeney et al 2005). Despite this, implants have been successfully placed in young children as young as three and a half years old with anodontia, and hypodontia of the anterior part of the mandible (Guckes et al 1997, Nunn et al 2003). This is because infraocclusion of the implant does not seem to be a problem in the former as there is no alveolar bone. With respect to the latter, studies have shown that positional changes are minimal in the anterior part of the mandible between the foramina, as growth occurs in the rami and the condyles (Bjork 1963, Nunn et al 2003) and also as a result of the closure of the midline suture in the first year of life (Meechan et al 2003). However, in contrast, the implant position is affected by sutural growth in the maxilla (Meechan et al 2003).

Implants are versatile in that the definitive restoration may incorporate crowns and for those with severe hypodontia or anodontia the provision of a stable overdenture(s) (Kearns et al 1999, Durstberger et al 1999). It has been reported

that the placement of implants promotes psychological wellbeing but has no effect on promoting craniofacial growth (Guckes et al 1997).

1.10.7.4 Autotransplantation.

Transplantation of a tooth from one part of the jaws to another in a subject is an alternative to implants and is a viable and useful alternative especially where there is hypodontia in one region of the mouth and crowding in another (Nunn et al 2003, Meechan et al 2003). Mandibular second premolars are a popular choice of tooth for transplantation into the maxillary anterior region, although canines and third molars have also been used (Kristerson 1985, Kristerson and Lagerstrom 1991, Meechan et al 2003). The success of transplantation is dependent on the amount of root formation with the optimum being between two-thirds to three-quarters. Failure due to resorption and loss of pulpal vitality increases with increased root formation. Furthermore, success is also dependent on minimal extra-alveolar time during the transplantation (Andreasen et al 1990a, 1990b). Nonetheless, teeth with mature apices can be transplanted but require root canal therapy about two to three weeks after the procedure (Meechan et al 2003). Transplanted teeth can be moved orthodontically if required without affecting root development (Kristerson and Lagerstrom 1991) and can be used as an abutment for a restoration, especially where several anterior teeth are missing (Jepson et al 2003).

1.10.7.5 Surgical correction of skeletal deformities.

Some subjects may require major maxillofacial surgery to correct skeletal discrepancy of either the maxilla and/or mandible in conjunction with implant provision with or without bone augumentation (Meechan et al 2003).

1.10.8 Maintenance and regular review.

Since the use of bridges carries the risk of debonding, caries and tooth movement the subject must be made aware of these risks and attend for follow up review and maintenance (Hobkirk et al 1995). Subjects' and their parents/guardians should be made aware that restorative treatment is not permanent and will lead to further restorative treatment in the future (Millar and Taylor 1995).

1.11 Self-esteem.

There are no research studies in the literature which have reported on the self-esteem of subjects with hypodontia, using a validated self-esteem inventory. Psychological upset and dissatisfaction with appearance have been reported in case reports of subjects with hypodontia (Jones et al 1978). The psychological literature suggests that there is a relationship between anatomical irregularities and ratings of self-esteem (Stricker 1970).

An individual's perception of the attractiveness of various aspects of his/her facial appearance appears to be influenced by their general self-esteem (Pitt and Korabik 1977). Furthermore the aforementioned authors, suggested that a person's perception of their facial profile is determine by their psychological self-satisfaction rather than by their objective appearance.

Evans and Shaw (1987) found in 50 prospective subjects for orthodontic treatment that self-ratings of dental attractiveness, using a scale referred to as the Standardised Continuum of Aesthetic Need (SCAN) and self-esteem, using Piers-Harris Children's Self-concept Scale, are associated. Further analysis of the over and under ratings for dental attractiveness and self-esteem found that over-rating dental attractiveness is not associated with high self-esteem, but under-rating of dental attractiveness is associated with low self-esteem.

From the previous literature the psychological impact of a craniofacial abnormality has been well documented. A heightened self-esteem has been reported by both Brantley and Clifford (1979a) and Leonard et al (1991) with subjects with cleft lip and/or palate abnormalities. In contrast Pope and Ward (1997) reported a low global self-esteem. Locker (1988) reported that psychological outcomes of oral conditions, for example, low self-esteem are ignored in terms of health as they impact on the individual, whereas loss of work because of caries is termed an important indicator of health as this, impacts on the economy and society.

Leonard et al (1991) further reported on the importance of the influence of parents in cultivating their child's development, of provision of understanding and support to their child in relation to the impairment. Additionally, these authors suggested that these children may have developed successes in other fields in their lives which reduced the psychological impact of the impairment. It is worthy, to note that the children and parents in the aforementioned authors study, had received professional intervention and the effect on both the parent and child's coping skills were unknown, but presumed to be of a positive nature. This highlights that cleft palate management over a period of years enables the subject to "overcome" a potentially handicapping condition (Brantley and Clifford 1979a, Leonard et al 1991).

1.12 Self-esteem, Self-concept, Self-image, and Self-perception.

The literature relating to self-esteem, self-concept, self-image, and self-perception is confusing. Certain terms are seldom adequately explained and often there is the assumption that the reader knows what they are (Gussy and Kilpatrick 2006).

1.12.1 Definitions and measurement.

In order to understand the exact meaning of these terms, definitions were sought.

A definition of:

- *self* is "what one is" (Chambers Twentieth Century Dictionary 1968).

- *Self-esteem*

  - Chambers Twentieth Century Dictionary (1968) defines self-esteem as having a good opinion of oneself.

  - Battle (2002) defines self-esteem as the attitude that an individual has towards him or herself.

  - The Webster II New College Dictionary (1995) defines self-esteem as an attitude of acceptance, approval, and respect towards oneself, manifested by personal recognition of one's abilities and achievements and an acknowledgement and acceptance of one's limitations (Battle 2002).

- *Self-concept* is defined as "the whole set of attitudes, opinions and cognitions that a person has of himself" (Collins English Dictionary 1982). (cognitions meaning "acquired knowledge including perception, intuition and reasoning")

- *Self-image* is defined "as one's conception of oneself (Oxford Compact Dictionary and Thesaurus 1997).

- No definition of *self-perception* was found in the dictionaries but "perception" is defined as "the act or faculty of perceiving" and according to the Thesaurus "perception" can be substituted for appreciation, awareness, sensation, view etc (Oxford Compact Dictionary and Thesaurus 1997).

In some literature, terms such as self-concept and self-esteem are being used synonymously whereas, in others, it has been reported that they are "two discrete dimensions" (King 1997, Battle 2002, Gussy and Kilpatrick 2006). With reference to the latter, self-concept was reported as being a description, for example curly hair, whereas self-esteem is an evaluation of the degree of satisfaction and therefore, can be considered to be positive, negative or neutral. However, it is generally accepted that with self-description, self-evaluation is often included spontaneously (King 1997, Gussy and Kilpatrick 2006). Listed overleaf, are other terms often associated with self-esteem/self-concept, which compound the confusion (King 1997).

- self-confidence
- self-acceptance
- self-appraisal
- self-worth
- self-satisfaction
- self-evaluation
- self-ideal
- self-love
- sense of adequacy
- personal efficacy
- sense of competence
- congruence
- ego
- ego-strength

Gussy and Kilpatrick (2006) reported that these factors may contribute to differences in findings in the literature. Furthermore, King (1997) reported that not only were there differences of opinions on the definitions but also on the measurement of self-concept and self-esteem. Table 1.11 overleaf shows some of the scales commonly used (King 1997).

Table 1.11 Scales used in the measurement of self-esteem and self-concept (King 1997).

Scales of Measurement	
Self-Concept	**Self-Esteem**
Multidimensional Self-Concept Scale (1992)	Coopersmith Self-Esteem Inventories (1987)
Piers-Harris Self-Concept Scale "The Way I feel About Myself" Scale (1984)	Culture-Free Self-Esteem Inventories, 2nd edition (1992)
Pyrt-Mendaglio Self-Perception Scale (1992)	Gordon Personal Profile (1978)
Self-Description Questionnaire I (1988)	Hare Self-Esteem Scale (1977)
Self-Description Questionnaire II (1990)	Robson Self-Esteem Questionnaire (1988)
Self-Perception Profile for Children (1988)	Rosenberg Self-Esteem Scale (1965)
Tennessee Self-Concept Scale (1988)	Self-Esteem Index (1991)

1.12.2 Overview of self-esteem, self-concept, self-image and self-perception.

Literature supports the fact that how individuals feel about themselves is paramount to their growth and development. Furthermore, positive self-esteem and realistic self-concepts are important factors in adopting healthy behaviour (King 1997). Psychological appreciation of oneself begins early in life, and how individuals view themselves affects their ability to relate to others and be accepted by others (Jones et al 1978). Indeed, Jokovic et al (2002) reported on the literature relating to age and the beginning of abstract thinking and self-concept. According to developmental psychology this appears to occur about six years of age. Children commence by comparing physical features and personality traits with their peers or against the norm. With age, their ability to evaluate appearance, emotions, relationships and behaviours develop, becoming complex and sophisticated around adolescence (Jokovic et al 2002). Sound psychological self-image is heavily influenced by the perceptions of our peers. As a result, for the child who looks different, for example those with hypodontia or cleft lip and palate, this may be a problem (Jones et al 1978). In fact, Burden and Pine (1995) found that peer group influences were more significant than social class or gender in the uptake of orthodontic treatment. Indeed, in their study of 506, 15-16 year old schoolchildren, using a single stage stratified random sampling technique, they found that children with similar dental aesthetics have similar perceptions of their malocclusion irrespective of their gender or social background. However, Shaw (1981b) found, in his study of 200 schoolchildren randomly selected from a school inspection programme, 100 of each gender, aged between 9-12 year olds, that dissatisfaction with dental appearance was commoner among the girls and was associated with increasing age. Girls were found to consider themselves of

below average attractiveness, more girls than boys admitted to frequent mirror viewing and more than twice as many girls as boys were dissatisfied with their dental appearance. Therefore, it is not surprising that twice as many girls as boys receive orthodontic treatment (Shaw et al 1979).

Dekovic and Meeus (1997) reported that a positive self-concept seemed to correspond to acceptance of the child by the parent(s) and a warm close parent-adolescent relationship. In this type of relationship, the adolescent is given support, positive reinforcement and love withdrawal is avoided in disciplining, which naturally promotes feelings of self-worth. Furthermore, the aforementioned authors reported that adolescents who had a more satisfying relationship with parents also had a more positive quality relationship with peers. However, Brantley and Clifford (1979b) reported that children with clefts perceived their parents as viewing them less favourably as compared to non-cleft children.

The importance of the home environment is highlighted by Emery et al (1993) who reported that the higher an individuals self-esteem in the parameters of not only home but also school the lower their use of substances such as smoking, alcohol and drugs.

Psychological well-being is dependent on a subject's perception of their facial attractiveness and their ability to communicate (Kapp-Simon and McGuire 1997). Turner et al (1997) reported that in their study of 112 subjects, aged 15-20 years old with craniofacial abnormalities, 73% perceived that the cleft had affected their self-confidence "very much" and 15% of parents felt that there was lowered self-

confidence due to the cleft. Furthermore, 100% of these subjects experienced communication problems with strangers for example initiating conversations or a job interview. The subjects felt not only self-conscious of their appearance, but sceptical of complimentary remarks attributed to their appearance.

1.13 Self-image.

1.13.1 The dimensions of self-image.

Simmons et al (1973) explained that self-image can be thought of as an attitude and therefore consists of different dimensions. Furthermore, they expanded in detail on four of these dimensions, namely, self-consciousness, stability of self-image, self-esteem and the perceived self.

- Simmons et al (1973) described *self-consciousness* as an individual's account of other's reactions to himself and his behaviour. The authors explained further that some people become involved with what they are doing and that concerns about how they are doing or what others think of them is not an issue. Whereas, for others, interaction becomes difficult and uncomfortable because issues of what others think or how they compare themselves to others is paramount.

- The second dimension of the *stability of self-image* is described by Simmons et al (1973) as a true concept of what you are like and uncertainty in this will lead to an inability to act or make decisions.

- The third dimension mentioned by Simmons et al (1973) is of *self-esteem* which is the individual's global positive or negative attitude towards him or herself. Global self-esteem is the individual's general feeling towards themselves (Simmons et al 1973). However an individual also has attitudes towards specific qualities such as looks, intelligence etc (Simmons et al 1973).

- The aforementioned authors believe as a result of other literature that the *"perceived self"* has an extremely important bearing on the self-image, particularly self-esteem.

Moreover, Simmons et al (1973) pose the thought that there is reason to think that changes in these dimensions would be disturbing or uncomfortable for the individual.

1.13.2 Self-image varies with age.

Using indexes developed to measure the four aforementioned aspects of self-image, Simmons et al (1973) found in their cross-sectional study of 1,917 school pupils, differences in different age groups. Early adolescents, particularly those around 12-13 years of age were shown to have increased self-consciousness, greater instability of their self-image, slightly lower global self-esteem, lower specific self-esteem and a less favourable view of themselves by others compared to children in the 8-11 year old group. In late adolescence, aged 15 years old, there was an improvement in self-consciousness, stability and global self-esteem.

However, no improvement was reported in their perceived self or specific qualities of intelligence, appearance, honesty, diligence and good behaviour.

The aforementioned authors suggested that, there is the possibility that the lower self-ratings on the specific qualities in the early adolescence compared to the younger age group, may be a reflection that adolescents are more "realistic" while younger children tend to "inflate" their self-qualities (Simmons et al 1973).

1.13.3 Self-image disturbance.

Simmons et al (1973) suggests that it is reasonable to assume that lowered self-evaluations indicate some degree of self-image disturbance, but this conclusion is not certain. To explain, the aforementioned authors write that a low self-rating on a quality which the individual values highly is likely to be experienced as disturbing, whereas, if the individual cares little for that particular quality they will not be upset by its lack. Therefore, if appearance is considered a quality that the individual values highly and unfortunately hypodontia is present, affecting the individual's facial appearance, disturbance in their self-image will occur.

1.13.4 Facial self-image.

Improvement in dental appearance rather than health and function is the main motivating factor for seeking orthodontic treatment (Tulloch et al 1984). Self-perception of dental appearance and perception of parents with regard to their offspring's facial and dental aesthetics are key factors in seeking orthodontic

treatment, especially if the irregularity is severe and/or visible (Kilpelainen et al 1993, Shaw 1981b).

1.13.5 Awareness facial self-image in relation to age.

Generally, awareness of facial self-image increases with age, peaking around the mid-teens (Shaw et al 1981b, 1991, Richman 1983, Kilpelainen et al 1993,). Younger children are less likely to be aware of their perception of their malocclusion (Horowitz et al 1971).

Hobkirk et al (1994) in their retrospective study of 451 subjects, aged at the time of their initial visit between 4-28 years, attending a Hypodontia Clinic over a ten year period, found that 40.1% of the subjects had no complaints. On closer analysis, it was found that this "no complaints" sample was comprised of younger subjects, with a mean age of 12.7 years. Hobkirk et al (1994) analysed the data regarding those subjects complaining about appearance (14.6%), which was defined as relating to the shape of teeth, the overall appearance of the arch and the jaw relationship, and found that in this "appearance" sample, the mean age was 17.7 years. However, it is noteworthy that some of those subjects complaining of missing teeth (20.7%) and spacing (16%) were not included in the "appearance" sample. From personal experience, subjects with hypodontia complaining about appearance, include missing teeth and spacing in addition to the others mentioned. The mean age of the children to include these two other parameters could not be ascertained. However, in this study, the peak age for first attendance at the Hypodontia Clinic is 13 years, which is in accordance to the aforementioned literature about increased awareness at this age.

Ingervall and Hedegård (1974) in a study of 18 year old males found a high awareness for certain anterior anomalies such as missing or malformed teeth but a low awareness for other aspects of malocclusion such as increased overjet, reverse overjet, anterior open bite or deep overbite. Interestingly, only 4% of the 287 sample considered themselves in need of orthodontic treatment although the objective need was determined by orthodontists to be 60%.

1.14 Attractiveness.

1.14.1 Physical attractiveness.

The mid 18th century proverb "beauty is in the eye of the beholder" is as true today as it was in the past. In today's society, people are judged constantly on the basis of their attractiveness, especially facial attractiveness (Cunningham 1999). Indeed, facial deformity and obesity were reported to be regarded as the least preferred trait (Richardson 1970). Any deviation from the individual's "ideal" and "perceived" body images can often result in discontentment, and lead to a desire to actively "normalise" the deviation, whether it be new clothes, new hair style, going on a diet or seeking orthodontic treatment (Shaw et al 1991). Indeed, physically attractive individuals are judged to be more socially desirable, secure more prestigious jobs, experience happier marriages, enjoy more fulfilling social and occupational lives and in all have more total happiness in their lives (Dion et al 1972).

1.14.2 Physical attractiveness and gender.

Lerner and Gellert (1969) found in his study of 45 pre-school children that girls were better than boys in comparing physical attractiveness. The author suggested that girls develop earlier awareness of physical differences in appearance, and that this possibly reflects adult expectations on the importance of attractiveness.

1.14.3 Physical attractiveness and age.

Very young children have been reported to have awareness of the characteristic of physical attractiveness (Dion and Berscheid 1974). The aforementioned authors pose a thought that although an individual's physical attractiveness may change with increasing age, early negative interactions associated with an unattractive appearance may be more difficult to overcome psychologically. Maybe this could be a problem for the subject with hypodontia who has undergone treatment to improve their appearance, but who in childhood had a late diagnosis, and/or had to wait for definitive treatment and was subject to negative interactions during this period of growing up. These thoughts fuel the importance of an early diagnosis so that treatment can reduce the effect of these possible negative peer interactions.

1.14.4 Physical attractiveness and popularity.

Dion and Berscheid (1974) also found that popularity among peers, especially in girls, increases with age and attractiveness. Attractive children generally tended to be considered to be more self-sufficient (capable of accomplishing what they wish) and independent (Dion and Berscheid 1974). Lowenstein (1978) reported that children who were popular, conforming to group norms and were socially

skilled, tended to be less bullied than those with the opposite traits. According to this study, it was observed that unattractive children are more likely to be victims of bullying (Lowenstein 1978).

1.14.5 Facial attractiveness.

There is some evidence that the oral region is of primary importance in determining facial attractiveness (Terry 1977). Facial preferences have changed throughout time from Palaeolithic man, to the Egyptians, to the Greeks, to the Romans, to the Renaissance to the present, and it is accepted that there is no ideal for facial appearance or attractiveness, and no equation can express the complexities of facial aesthetics (Peck and Peck 1970). The mass media in the form of television, films, newspapers and magazines are very influential in unifying people's attitudes to facial attractiveness (Peck and Peck 1970, Shaw 1981c). The area around the mouth is used in speech, eating and mirroring emotions, and thus has great importance in both social interaction and self-image. Therefore, any deviation from the normal is highly visible and will interfere with our interaction with others, and unless there is functional disability, the handicap is tragically social and psychological (MacGregor 1970, Shaw 1981c). Indeed, Shaw (1981c) reports historically on the misconceptions associated with facial deformity. Tobiasen (1987) reported that irrespective of age or gender, deformed faces were regarded as less friendly, less popular, less likely choices as friends, less smart, and less good-looking than non-deformed faces.

It is generally agreed that the most preferred appearance is that of the ideal incisal alignment (Shaw et al 1980, Shaw 1981a, Tulloch et al 1984). Horowitz et al

(1971) reported that there was generally uniform agreement of the hierarchy of preferences for given occlusal relations, regardless of race. The preferential order from the highest to the lowest was:

- Ideal occlusion
- Anterior open bite
- Mandibular protrusion
- Midline deviation
- Maxillary protrusion
- Excess spacing
- Bi-maxillary protrusion
- Crowding
- Repaired cleft lip

1.14.6 Facial attractiveness and gender.

Dentofacial anomalies have been shown to detracted from the attractiveness of the faces of girls more than boys (Shaw 1981a, Tobiasen 1987).

1.14.7 Cultural influences of facial attraction.

Although self-perception is a deeply personal matter, similar perceptions can be explained by commonly held views of attractiveness within a culture or society (Albino and Tedesco 1991). Indeed, Shaw (1981c) reported that facial deformity results in different prejudices in different cultures.

1.15 Social interaction.

1.15.1 Physical attractiveness and social interaction.

Recent research shows that physical attractiveness does affect other's reactions, especially those of peers (Miller 1970, Dion et al 1972). Attractive persons were perceived to possess more socially desirable personalities such as friendlier, warmer, more stable and more sincere compared to unattractive individuals (Dion et al 1972). Interestingly, research has shown that children's ratings of physical attractiveness do not differ significantly from those of adults (Dion 1973, Cross and Cross 1971). Presumably, this is a result of cultural standards concerning aspects of attractiveness being transmitted either directly or indirectly from adults to children (Dion and Berscheid 1974).

Kleck et al (1974) reported that friendship choices and being liked (social acceptance) occurred more often when a subject was attractive. Interestingly, facial cues were identified as being important in making the attractiveness decision. The face (73%) was referred to by the majority of respondents, with specific features such as the hair (53%), the eyes (47%), the teeth (47%), the mouth (40%), the lips (26%) and ears (20%) being identified.

1.15.2 Facial attractiveness and social interaction.

Facial attractiveness influences social interactions (Faure et al 2002). There is an assumption that people with a severe orthodontic Class II or Class III malocclusion are "slow" and "dull" (Peck and Peck 1970). Moreover, those with Class III malocclusions are perceived to be aggressive (Shaw 1981c). Children

with a normal dental appearance were judged to be better looking, more desirable as friends, more intelligent and less likely to behave aggressively (Shaw 1981a).

Interestingly, it was found that subjects with severe facial deformities which evoked either pity or revulsion were psychologically in better shape than those with lesser deformities. An important factor is that those with severe facial deformity expect a negative response wherever he/she goes and consequently develop skills to cope with this. However, for those with a less obvious deformity, of which orthodontic malocclusions are included, the response was inconsistent and unpredictable. This can give rise to anxiety in the individual who was subject to ridicule. Derisive laughter is destructive and the shame, anger and distress it can cause to individuals is immeasurable (MacGregor 1970). Furthermore, Kapp-Simon and McGuire (1997) in an observational study reported that adolescents with a craniofacial condition behaved differently to non-affected peers in a natural, daily occurring situation. Interestingly, about half of the subjects with craniofacial condition who were approached to take part in the study declined. Indeed, it was noted that those who declined were those deemed by the clinical staff to be in need of the service of the social skills group. Those with the craniofacial condition were observed to be passive rather than active participants in conversations. They were less likely to initiate a conversation and less likely to respond to a peer if approached. On the other hand, non-affected peers were less likely to respond to affected peers if approached, and furthermore, addressed affected peers at a less frequent rate. This observational study only considered the frequency of interaction and not the quality of the interaction. Kapp-Simon and McGuire (1997) further reported that some children in this situation feel that it is

safer to have limited interaction with peers as they are able to avoid uncomfortable interactions.

The act of repeated comments about a physical or social characteristic in order to make fun of them is "teasing". Turner et al (1997) reported that, out of a sample of 112 with a craniofacial abnormality, 60% were teased regarding their speech and appearance and in 96% of those cases, the focus of the tease was the cleft. Of a more malicious nature, is harassment. This involves verbal and physical intimidation (Shaw et al 1980). Shaw et al (1980) found in their study of 531 schoolchildren aged from nine to thirteen years old, from six different schools that, 66% were teased about one or more characteristics, which included height, weight, hair, teeth, glasses, clothes, freckles, strength etc. With reference to harassment, 26.5% suffered verbal intimidation compared to 14.5% who suffered either physical intimidation or other methods of victimisation. Seven per cent (37 children) were regularly teased about the appearance of teeth. It is worthy to note that of these 37 children, 19 were related to prominence of incisal teeth, 3 regarded crowding and the remaining 15 were due to a fractured tooth, and attributed to poor oral hygiene or caries. Researchers confirmed that 30 out of the 37 children did have a malocclusion. However, teasing due to spacing which may be attributed to possible hypodontia was not reported in this study. A possible reason for this is that, in this age group, the normal stage of dental developmental has natural spacing. However, normally with increasing age and further dental development, this natural spacing will decrease or close completely. Of those with hypodontia, often these spaces persist and can in some cases worsen with further exfoliation of primary teeth. As a result, spacing in the anterior region will

become especially obvious not only for the individual, but to observers. Teasing about teeth is known to be the most hurtful (Shaw et al 1980). In fact, not only is teasing of dental features twice as likely to lead to general harassment than those not teased about their teeth but also those school pupils who had complained of harassment were more likely to be labelled by their teachers as "introverted". The authors suggested that, other features and qualities of the individual are likely to heighten or decrease the "teasible feature". Indeed, the authors suggested that sustained ridicule and insult may predispose to lower self confidence and alienation (Shaw et al 1980).

1.16 Relationship between severity of the dental defect and psychological impact.

Research into the impact of hypodontia is scanty. Hypodontia is either omitted completely or if mentioned, it is incorporated under malocclusions. Much of the research, in dentistry, into psychological impact has involved orthodontic malocclusions or those with orofacial defects (mainly cleft lip and palate). A facial disfigurement or speech defect is easily detectable, whereas psychological adjustment to a disability is more difficult to recognise (Richman and Eliason 1982).

1.16.1 Subjects with an orthodontic malocclusion.

According to Shaw (1981b) only a minority of malocclusions can be regarded as seriously handicapping and although he does not specify which, it is accepted that hypodontia is in this category. However, Jones et al (1978) describe severe

hypodontia as physically and psychologically crippling. Shaw (1981b) reported that children with severe visible irregularities not only had more negative feelings about their teeth but they, and their parents also judged the teeth to be the worst feature. The judgement of the severity of the irregularity appeared to be based on the size of the overjet. Hypodontia was not specifically mentioned except in respect to the adverse appearance when spacing is present. However, in this age group such spacing could be attributable to normal development. In addition, it was suggested that satisfaction with dental appearance and desire for orthodontic treatment were not affected by social class, intelligence and general self-esteem. However, the self-esteem measurement in this study was a superficial estimate using a seven point scale on the basis of one question, rather than a validated questionnaire (Shaw 1981b).

Kilpelainen et al (1993) confirmed that an increased overjet was a significant predictor for a desire for orthodontic treatment for an improvement in facial appearance and that early treatment and even partial correction of the malocclusion may have significant psychological benefits for a sensitive child. Despite this, Albino et al (1994) reported that in subjects with mild to moderate malocclusions, treatment improved parent, peer and self-evaluations of dento-facial attractiveness but the subject's self-esteem was unaffected. The aforementioned authors found this a surprising finding. They speculated that this particular aspect of facial aesthetics may be too insignificant to affect self-esteem or adolescents may be able to evaluate their dento-facial appearance independently, without incorporating these judgments into their self-esteem. Furthermore, Albino et al (1994) suggested that perhaps the self-esteem of

adolescents is more related to interpersonal performance than either facial or physical attractiveness.

1.16.2 Subjects with cleft/lip palate.

Pope and Ward (1997), Richman (1983) and Richman et al (1985) reported that adolescents with cleft lip and palate, who reported social adjustment problems, were concerned with their facial appearance. In fact, Pope and Ward (1997) reported that their dissatisfaction with appearance was associated with low global self-esteem. Despite, Kapp (1979) in his study also reporting that adolescents with clefts were more dissatisfied with their appearance than non-cleft children, he found that there was no difference in respect of global self-concept scores. Furthermore, girls with clefts reported more anxiety, less success in school and more unhappiness and dissatisfaction with their appearance compared to their non-cleft peers.

In the study by Richman et al (1985), the subjects reported an unrealistic perception of their facial appearance: They rated their appearance better than independent judges. It thought that this denial of facial disfigurement is a defence mechanism that results in social inhibition.

Leonard et al (1991) reported that in their study of subjects with cleft lip and palate, 98% of the children reported above average or average self-concept using the Piers-Harris Children's Self-Concept Scale. This was a surprising result, as a negative self-concept was anticipated. However, adolescent girls showed a more negative self-concept compared to younger girls. Like Kapp (1979), physical

appearance was identified as an area of concern for these subjects. Furthermore, Kapp-Simon et al (1992) found in their study of adolescents with cleft lip and palate abnormalities that their self-esteem was not poor but rather that, they were at risk of psychological adjustment. Kapp-Simon et al (1992) and Leonard et al (1991) both report that these affected subjects make psychological adjustments with a degree of inhibition, in other words children will withdraw from the social scene in order to reduce their distress and preserve their self-esteem. However, Kapp-Simon et al (1992), further felt that self-concept scores may not be the best indicators of overall psychological functioning and therefore, to identify those of psychological risk, use of a well validated measure of adjustment would be more appropriate.

Richman (1983) suggested that dissatisfaction of facial appearance with cleft lip and/or palate subjects combined with the expectation or hope of facial surgery, creates a continuation of self dissatisfaction. The danger of the expectations of the achievement of cosmetic enhancement have to be carefully assessed and explained to prevent the subject having unrealistic expectations (Richman 1983, Kiyak et al 1982).

1.16.3 Subjects with hypodontia.

It has been reported in case studies children with moderate to severe hypodontia tend to be increasingly self-conscious, withdrawn and lacking in self-confidence, especially in adolescence. Often as a result of a lack of dentition which obviously deviates from the ideal they may suffer ridicule and rejection from their peers and find socialising difficult (Duggal and Ogden 1990). There is no literature using a

validated and published scale of measurement, to evaluate the impact of hypodontia on subjects, especially with specific respect to self-esteem.

1.16.4 Quality of life.

Locker (1988) reported on the literature of the effects of disease and disability. Indeed, in specific relation to dental and oral disease, this is often related to physical attractiveness. Locker et al (2002) explored the impact on the family of children with oral and oro-facial conditions. Hypodontia was included under the malocclusions in the orthodontic group. The impact in the orthodontic group was financial whereas, in the oro-facial group, it was reported to involve time off work for parent(s)/guardian(s), parental feelings of guilt, concern of the future for the child and interfamilial jealousy.

Furthermore, a comparative study by Locker et al (2005) of the quality of life of those aged 11-14 years with orofacial conditions compared to those with dental caries, using the health-related quality of life (HRQOL) questionnaire found that, there was no difference between these two groups. The conclusion from this study was that despite difficulties, such as, of being teased, asked questions, left out by other children, arguing with others regarding their condition, the overall quality of life of those with an orofacial condition was no different from that of those with a common oral condition. A criticism of this study may be that the comparison was with a group with an oral condition rather than a control group without any oral condition. This study also did not measure satisfaction with appearance, and it is assumed that there is no difference in the two groups in feelings of attractiveness on the basis of the responses to questions in the emotional well-being domain.

Locker et al (2005) also reported that the outcomes of well-being and quality of life are a result of an interaction of the condition and the availability of material, social and psychological factors, which allow the subject and/or family to cope with the challenge of the condition presented to them.

1.17 Summary of literature review.

Hypodontia is one of the most common dental anomalies, occurring more frequently in the permanent dentition compared to the primary dentition. The prevalence rate for the primary dentition is considered to be in the range 0.1-0.9% and in the permanent dentition 3.5-6.5%, excluding third molars. The prevalence of hypodontia of the third permanent molar has been reported in the range of 25-27%. It is generally accepted that the most common missing teeth, in descending order is: third molars, mandibular second premolars, maxillary lateral incisors and second premolars. There is a gender predilection in that, females are more affected by hypodontia than males, showing a female : male ratio of 3 : 2. There are race differences, with Caucasians being more affected than African Americans. Hypodontia is associated with other dental features. The effects of hypodontia can be profound and corrective treatment complex requiring the expertise of a multidisciplinary team.

Hypodontia can occur in association with genetic syndromes, or an isolated sporadic or familial trait. The mode of inheritance for familial hypodontia is generally accepted as an autosomal dominant pattern with incomplete penetrance and variability expressivity. Genetic markers for familial hypodontia, Msx 1 and Pax 9 have been identified. There is a possibility that another gene, transforming growth factor alpha (TGFA) may also be involved.

Attractive individuals are perceived to be more socially acceptable and, in general, lead more successful lives. The presence and the ideal incisal alignment

of teeth contribute to facial aesthetics. Aesthetics rather than function is the main motivating factor for seeking orthodontic treatment, especially with females. It has been well documented that subjects with a craniofacial abnormality or an obvious malocclusion have been subjected to "teasing". At present there have been no reports on the aspect of "teasing" of subjects with hypodontia. Well-being and the quality of life are dependent on coping with the challenges of a condition through social and psychological means and also through material means; for example financial or accessing services. Self-esteem is having a good opinion of one's self and how one perceives one's self (self-image) is often associated with appearance. Research involving appearance and orthodontic treatment requests, the impact of facial deformity, and the appearance and self-esteem of cleft lip and palate subjects is well documented. However, research of the impact of appearance in association with self-esteem of subjects with hypodontia is non-existent and therefore, long overdue.

Chapter 2 Aims.

2.1 Aims.

- To report on the characteristics of subjects attending the Hypodontia Clinic at Glasgow Dental Hospital and School, with particular reference to the number and the pattern of missing teeth, and treatment needs.

- To report on the level of self-esteem in adolescents with hypodontia compared to matched controls.

2.2 Null hypothesis.

There is no difference in self-esteem of subjects with hypodontia compared to non-hypodontia controls.

Chapter 3 Material and Method.

3.1 The study design.

The prospective study incorporated two parts;

Part 1: The characterisation of the subjects, with particular reference to the number and pattern of missing teeth, and treatment need who attended the Hypodontia Clinic at Glasgow Dental Hospital and School from May 2005 to May 2006, using data collected from a hypodontia clinical assessment form.

Part 2: A prospective controlled study which evaluated the self-esteem of a sample of adolescents with hypodontia, who attended the Hypodontia Clinic at Glasgow Dental Hospital and School from May 2005 to May 2006.

3.2 Part 1: The characterisation of subjects referred to the Hypodontia Clinic.

3.2.1 Ethical approval.

Ethical approval had already been obtained by Dr M.T. Hosey from the Area Dental Ethics Committee to establish a database for all consenting subjects who attended the Hypodontia Clinic at the Glasgow Dental Hospital and School (see Appendix 1).

3.2.2 Subject selection and recruitment.

Recruitment of participants occurred when they and their parent/guardian, attended the Hypodontia Clinic. It was intended that all subjects and their parents/guardians, who attended the Hypodontia Clinic be approached, informed of the study and recruited by the post-graduate (MSc) research student, Mrs Rosemary Broad. Both the subject (usually a child) and parent/guardian were verbally informed and permission gained regarding the data collection of their assessment examination findings. An information sheet was issued (see Appendix 2) and a signed consent form gained from those who participated (see Appendix 3).

3.2.3 Data collection.

The data was encoded to ensure anonymity. The following data was recorded on a clinical assessment form (see Appendix 4 and 5) : age, gender, race, partial post

code, charting of teeth present, past dental history was recorded with specific reference to any permanent tooth extractions, the number and type of missing teeth, pattern of missing teeth, family history of missing teeth (pedigree), orthodontic malocclusion, overjet, spacing, other clinical observations, whether the subject had a presenting complaint, treatment and speciality involvement. The partial post code was used to determine the level of social deprivation using the Carstairs Index (DEPCAT score) (McLoone 1994). The subject's treatment need was recorded, for example space closure or opening, camouflaging in form of composite build ups, veneers, bridges, partial/over dentures or implants.

Where possible the subjects were categorised by the pattern of missing permanent teeth and possible gene markers, as follows:

(1) Maxillary lateral incisors only (unknown gene/perhaps TGFA)

(2) Second premolars and third molars (Msx 1)

(3) Molars (Pax 9m)

(4) Molars, second premolars and mandibular central incisors (Pax 9pi)

(5) Molar and incisor (unknown gene/possible TGFA involvement)

(6) Combination of incisors (unknown gene/perhaps TGFA)

(7) Canines either alone (unknown gene); or with third molars (unknown gene); or with third molars and incisors (unknown gene/possible TGFA involvement)

(8) Premolars (unknown gene)

(9) Premolars and incisors (unknown gene/possible TGFA involvement)

(10) Severe hypodontia of molars and premolars with other teeth

(possible undiagnosed syndromes)

Recording of the data on the clinical assessment form was undertaken by the various examining dentists attending the Hypodontia Clinic. There was no calibration of the examining dentists undertaken. Collection and subsequent inclusion of the data into the hypodontia database was performed by the post-graduate (MSc) research student, Mrs Rosemary Broad who rechecked, where possible, the charting by referring to the radiographs. The database was specifically developed for the hypodontia clinic by Mrs Rosemary Broad,

3.2.4 Statistical analysis.

The results are presented by descriptive statistics.

3.3 Part 2: A prospective controlled study to evaluate the self-esteem of a sample of adolescents with hypodontia.

3.3.1 Ethical approval.

Further ethical approval was gained from the West Glasgow Ethics Committee II and North Glasgow University Hospital Research and Development by the post-graduate (MSc) research student, Mrs Rosemary Broad for part 2 of the study (see Appendix 6).

3.3.2 Recruitment.

Subjects aged between 13 and 18 years, with hypodontia, who attended the Hypodontia Clinic were invited to participate as the study group.

Subjects, aged between 13 and 18 years who attended the Community Dental Clinic at Clydebank Health Centre for routine dental treatment were invited to participate as the control group.

In both the study and control groups, recruitment was performed by the post-graduate (MSc) research student, Mrs Rosemary Broad. The recruits and their parent/guardian were informed both verbally and by information sheet (see Appendix 7), and thereafter, informed consent was obtained (see Appendix 8).

A target of 30 subjects in each group was set, as this was the number of recruits in previous, similar studies (Cornman 1993, Bolden and Williams 1995, Iniewicz 2005).

Moreover, a feasibility study, based on an audit of the Hypodontia Clinic database determined this to be a pragmatic number of potential hypodontia recruits, in the age range, during the time period of the study. It was intended to perform a statistical power calculation to test the adequacy of the sample size.

3.3.3 Selection Criteria.

3.3.3.1 The hypodontia study group:

- subjects aged 13-18 years old with hypodontia.
- basic English skills and literate.

Subjects who had a known (diagnosed) syndrome associated with hypodontia, for example, Down's syndrome or Ectodermal dysplasia were excluded. However, it was accepted that the sample might include those with undiagnosed syndromes, particularly ectodermal dysplasia.

Subjects who were undergoing treatment for their hypodontia were also excluded.

3.3.3.2 The control group:

- subjects, aged 13-18 years old without hypodontia.
- basic English skills and literate.

Subjects undergoing orthodontic treatment were excluded, as it was deemed probable that they would answer the self-esteem questionnaire with regard to the wearing of an orthodontic appliance.

3.3.4 Materials.

Two self-reporting questionnaires were used:

- Hosey-Bradnock questionnaire
- Culture-Free Self-Esteem Inventory

Both questionnaires were issued to the hypodontia study group but only the Culture-Free Self-Esteem Inventory (Battle 2002) was issued to the control group. The subjects were asked to complete the questionnaires on their own but with the support of the researcher (Mrs Rosemary Broad) as required.

3.3.5 Hosey-Bradnock questionnaire.

The Hosey-Bradnock questionnaire was developed by Dr M.T. Hosey and Dr G. Bradnock and had been previously piloted in the Hypodontia Clinic, at Birmingham Dental Hospital and School. It is an unpublished questionnaire to ascertain how subjects with hypodontia felt about their teeth, the depth of feeling,

their attitude towards wearing orthodontic appliances and dentures, and what treatment they desired. It is shown in Appendix 9.

3.3.6 Culture-Free Self-Esteem Inventory.

The responses were recorded on the adolescent student response form of the Culture-Free Self-Esteem Inventory (see Appendix 12). These responses were analysed on the profile/scoring form of the Culture-Free Self-Esteem Inventory (see Appendix 13). The data recorded from the profile/scoring form of the Culture-Free Self-Esteem Inventory was as follows:

- Global Self-Esteem Quotient (GSEQ):- a numerical value of self-esteem.
- Defensiveness Score:- indicates integrity of the subject.
- Standard Scores for subscales:- differentiation of Global self-esteem into parameters of:
  - Academic self-esteem
  - General self-esteem
  - Parental/Home self-esteem
  - Social self-esteem
  - Personal self-esteem
- Descriptive Ratings:- a corresponding description of self-esteem to the GSEQ and Standard Scores for the subscales.

3.3.6.1 Evaluation of self-esteem using the Global Self-Esteem Quotient.

Scoring from the 67 responses of the Culture-Free Self-Esteem Inventory resulted in a numerical value for the Global Self-Esteem Quotient (GSEQ). This numerical value for the GSEQ represented a person's performance over the whole inventory and ranged from 38-161. Ranges of these numerical values for GSEQ corresponded to descriptive ratings of self-esteem. These are shown in Table 3.1.

Global Self-Esteem Quotients from 90-110 are considered **normal** or **average** self-esteem.

Table 3.1 Numerical GSEQ value ranges and the corresponding descriptive rating of self-esteem.

Global Self-Esteem Quotient (GSEQ)	**Descriptive Ratings**
>130	*Very high self-esteem*
121-130	*High self-esteem*
111-120	*Above average self-esteem*
90-110	*Average self-esteem*
80-89	*Below average self-esteem*
70-79	*Low self-esteem*
<70	*Very low self-esteem*

3.3.6.2 The Defensiveness Score.

The Defensiveness Score was not a measure of self-esteem; it was essentially a 'lie detector'. It was a measure of how willing the individual was to disclose socially unacceptable or undesirable behaviours, for example, "Have you ever taken anything that didn't belong to you?". There were eight questions which assessed this and the "cut off" score was four out of eight (see Appendix 14). Therefore, a score of four or more questioned the validity of the subject's self-esteem score, the GSEQ (see Appendix 15). However, it did not alter the Global Self-Esteem Quotient. Therefore, the GSEQ values from subjects with a Defensiveness Score of four or more were excluded from the later statistical analysis of the comparison between the two groups.

3.3.6.3 Standard Scores for the subscales.

The Culture-Free Self-Esteem Inventory is age specific. For the purpose of this study, the adolescent inventory for ages 13-18 years was used. The Culture-Free Self-Esteem Inventory was designed to take account of the evolvement of self-esteem with age. So, in the adolescent sample, as well as a Global Self-Esteem Quotient there was a further subcategorisation into Standard Scores for the subscales. These subscales were:

- Academic
- General
- Parental/Home
- Social
- Personal

The scoring from the responses resulted in a standard score for each of the subscales, which ranged from 1-20. Similar to the GSEQ, numerical ranges corresponded to descriptive ratings of self-esteem, which is shown below in Table 3.2.

Table 3.2 Numerical Standard Score of subscale value ranges and the corresponding descriptive rating of self-esteem.

Standard Score of Subscale	**Descriptive Ratings**
17-20	*Very high self-esteem*
15-16	*High self-esteem*
13-14	*Above average self-esteem*
8-12	*Average self-esteem*
6-7	*Below average self-esteem*
4-5	*Low self-esteem*
1-3	*Very low self-esteem*

Although the GSEQ was the most useful value derived, the Standard Scores for the subscales embellished this value as they indicated the person's strengths and weaknesses.

3.3.7 Statistical analysis.

Descriptive statistics were used to present the Hosey-Bradnock questionnaire results, GSEQ values, the Standard Scores of the subscales and the Defensiveness Scores.

The GSEQ values from both the hypodontia study group and the control group were found to be not normally distributed, so the results were summarised using medians and ranges. Furthermore, the Mann-Whitney test was used to compare the two groups. A level of $p < 0.05$ was determined to be significant.

Chapter 4 Results.

4.1 Part 1: The profile of subjects at the Hypodontia Clinic at Glasgow Dental Hospital and School.

4.1.1 Sample.

Of the 85 subjects who attended the Hypodontia Clinic, 71 were approached and, all accepted the invitation to participate in the compilation of the hypodontia database.

4.1.1.1 Age.

The age of the subjects ranged from 7 to 35 years with a mean age of 13.76 years and a modal age of 12 years. Thirty nine were aged 13-18 years. The age at presentation at the Hypodontia Clinic, is detailed in Table 4.1.

Table 4.1 Age at presentation at the Hypodontia Clinic at Glasgow Dental Hospital and School.

Age at presentation at the Hypodontia Clinic (years)	Number of subjects (Total n=71)
7-10	n=10 (14.1%)
11	n=5 (7.0%)
12	n=13 (18.3%)
13	n=11 (15.5%)
14	n=9 (12.7%)
15	n=11 (15.5%)
16	n=5 (7.0%)
17	n=3 (4.2%)
18	n=0 (0%)
19	n=1(1.4%)
27	n=1 (1.4%)
32	n=1 (1.4%)
35	n=1 (1.4%)

4.1.1.2 Gender and race.

Of the 71 participants, females accounted for 43 and males for 28. This represents 61% and 39% respectively of the total sample. All subjects were Caucasian.

4.1.1.3 Deprivation.

The distribution of the participants in respect to the level of social deprivation is shown in Table 4.2. Eleven participants were categorised as DEPCAT 1 and 2, the most affluent areas, whereas in DEPCAT 7, the most socially deprived area, there were eighteen participants.

Table 4.2 Distribution of participants in respect of DEPCAT categories.

Carstairs Deprivation Category Index (DEPCAT)	Number of participants (Total n=71)
1	n=2 (2.8%)
2	n=9 (12.7%)
3	n=11 (15.5%)
4	n=13 (18.3%)
5	n=14 (19.7%)
6	n=4 (5.6%)
7	n=18 (25.4%)

4.1.2 Family history of hypodontia.

A positive family history of hypodontia was reported by 30 (42%) of the participants whereas there was a negative response in 35 (49%). Furthermore, one (2%) was unsure, and for five (7%), the family history was not recorded.

4.1.3 Presenting complaint at the Hypodontia Clinic.

The presenting complaint of 48 (68%) of the participants was related to appearance of the teeth. These presenting complaints are further detailed in Appendix 11.

4.1.4 Clinical observations.

4.1.4.1 Reduction in tooth shape and size.

4.1.4.1.1 Microdontia.

Three subjects were reported to have microdontia.

4.1.4.1.2 Peg-shaped laterals.

Twelve maxillary lateral incisors were recorded as being peg-shaped. This occurred in seven subjects;

- Bilaterally in five subjects
- Unilaterally in two subjects
    - In one subject, the other maxillary lateral incisor was congenitally missing and in the second subject a dens in dente was present.

4.1.4.1.3 Taurodontism.

One subject was reported to have taurodontism and this affected all the first permanent molars.

4.1.4.2 Infraocclusion of primary molars.

Nine subjects presented with infraocclusion of primary molars. This predominately affected the second primary molars (n=14) compared to the first primary molars (n=2).

4.1.4.3 Enamel hypoplasia.

Five subjects were reported as presenting with enamel hypoplasia, although in one subject this was thought to have occurred as a result of trauma to the associated primary tooth.

4.1.4.4 Malocclusion associated with canines.

Palatally impacted canines were not reported in any of our subjects. Transposition of a maxillary canine with a first premolar was reported in only one subject. The anticipated hypodontia of maxillary lateral incisors associated with this type of transposition was not present.

4.1.4.5 Incisal relationship, skeletal pattern and teeth alignment.

Further clinical observations are detailed in Table 4.3 overleaf.

Table 4.3 Clinical observations.

Clinical observations		Number of sample Total n=71	Not recorded
Incisal Relationship	Class I	n=26 (36.6%)	n=23 (32.4%)
	Class II Div I	n=6 (8.5%)	
	Class II Div II	n=10 (14.1%)	
	Class III	n=6 (8.5%)	
	Normal overjet (2-4mm)	n=19 (26.8%)	20 (28.2%)
	Increased overjet (>4mm)	n=7 (9.9%)	
	Decreased overjet (<2mm)	n=23 (32.4%)	
	Unable to assess	n=2 (2.8%)	
Skeletal Pattern	Class I	n=26 (36.6%)	n=8 (11.3%)
	Class II	n=23 (32.4%)	
	Class III	n=12 (16.9%)	
	Class III / I (not clear if Class III or I)	n=2 (2.8%)	
	Crowding upper arch	n=6 (8.5%)	n=18 (25.3%)
	Spacing upper arch	n=32 (45.1%)	
	Crowding lower arch	n=4 (5.6%)	n=19 (26.8%)
	Spacing lower arch	n=24 (33.8%)	

4.1.5 Extracted teeth.

- Twelve subjects had 16 permanent teeth extracted.
- Fourteen teeth were extracted for carious reasons.
- Two teeth were extracted for orthodontic reasons.
- Two subjects had teeth charted as "not recorded" with a report of the possibility of extractions.

4.1.6 Congenitally missing teeth.

- The total number of congenitally missing teeth in the 71 subjects was 604.
- The number of congenitally missing teeth (excluding third molars) in this sample was 453.
- The total number of "not recorded" teeth was 146.
- The maximum number of congenitally missing teeth in a subject, (excluding third molars) was 23 teeth.
- The minimum number of congenitally missing teeth, in a subject, (excluding third molars) was one tooth.
- The mean number of missing teeth (excluding third molars) was six teeth. This is further detailed in Table 4.4 overleaf.
- 38 (54%) subjects had six or more congenitally missing teeth

Table 4.4 Details of the most commonly recorded missing teeth.

	Max. third molars	Max. second premolars	Max. canines *	Max. laterals incisors	Mand. canines *	Mand. second premolars	Mand. third molars
Congenitally missing	n=73	n=70	n=28	n=84	n=14	n=78	n=78
Not recorded	n=37	n=2	n=7	n=4	n=6	n=5	n=38

*There were eighteen subjects in total with missing canines.

4.1.7 Patterns of missing teeth.

Data was complete in 67 of the 71 subjects to enable interpretation of patterns of congenitally missing teeth. The data was considered void in four subjects because too many teeth were "unrecorded" in those subjects.

Table 4.5 overleaf shows the number of subjects in relation to the pattern of missing teeth and the possible gene marker. The gene TGFA has been included despite there being only "a borderline association in cases with at least one missing incisor" (Vieira et al 2004).

Table 4.5 Patterns of congenitally missing teeth

Category	Pattern of missing teeth	Number of subjects (Total n=67)
1	Maxillary lateral incisors	n=5 (7.5%)
2	Second premolars and third molars	n=2 (3%)
3	Molars	n=1 (1.5%)
4	Molars, second premolars and mandibular central incisors	n=0 (0%)
5	Molars and incisors	n=10 (14.9%)
6	Combination of incisors	n=1 (1.5%)
7	Canines alone (n=1) Canines and third molars (n=1) Canines, third molars and incisors (n=1)	n=3 (4.5%)
8	Premolars only	n=5 (7.5%)
9	Premolars and incisors	n=6 (9%)
10	Others all which have severe hypodontia (including molars and premolars)	n=34 (50.7%)

4.1.7.1 Further interpretation of the patterns within the sample (n=67): Extended patterns.

4.1.7.1.1 Maxillary lateral incisors.

A total of 47 subjects presented with congenitally missing maxillary lateral incisors either alone or in combination with other missing teeth. This accounted for 70% of the sample. Forty two subjects had missing maxillary lateral incisors within a pattern of other missing teeth. Five subjects presented with only maxillary lateral incisors missing; three bilaterally and two unilaterally.

4.1.7.1.2 Lower incisors.

Eighteen of the 67 subjects had lower incisors congenitally missing. There was only one subjects who had hypodontia of lower incisors and maxillary laterals alone. The remaining seventeen showed an extended pattern of further hypodontia to include molar and premolar involvement. Of these seventeen, sixteen extended the pattern to include maxillary laterals. Of these sixteen, six extended the pattern of hypodontia to include canines.

4.1.7.1.3 Canines.

Over a quarter of this sample were found to have congenitally missing canines. This was elaborated further in Table 4.6 overleaf.

Table 4.6 The patterns of missing canines

Number of subjects Total n=67	Pattern of congenitally missing canines
n=18 (26.8%)	Canines missing
n=17 (25.4%)	Maxillary canines missing
n=6 (9%)	Bilateral maxillary canines missing
n=5 (7.5%)	All maxillary and mandibular canines missing
n=1 (1.5%)	Only lower canines missing

Seventeen subjects had congenitally missing maxillary canines, twelve of these subjects were additionally missing the maxillary lateral incisor. This occurred bilaterally in ten subjects.

There was one set of siblings who both had congenitally missing canines within the sample. The elder brother had 11 congenitally missing teeth in total, which included a unilateral maxillary canine. His sister had 12 congenitally missing teeth in total, which included bilateral maxillary canines. Two other female subjects with congenitally missing canines reported a history of hypodontia in sibling sisters. In one, the congenitally missing teeth were not known, however, in the other the pattern of missing teeth had not included canines.

4.1.8 Accuracy of the dental charting.

The charting was deemed inaccurate if even one tooth was "not recorded". The initial assessment form was amended as the layout lead to charting inaccuracies. As a result of this only four chartings were deemed void. The improvement in charting accuracy is shown in Table 4.7 below.

Table 4.7 Quality assurance evaluation of the assessment form.

Total number of assessment forms (n=71)	**Accurate charting**	**Inaccurate charting**	**Possible mis-chart**
Initial assessment form (n=35)	n=16 (45.7%)	n=18 (51.4%)	n=1 (2.9%)
Amended assessment form (n=36)	n=32 (88.9%)	n=4 (11.1%)	n=0 (0%)

4.1.9 Treatment and speciality involvement.

Of the database sample 51 (72%) were subsequently referred for complex integrated care involving two or more specialised departments. Three (4%) were monitored for further dental development pending future treatment and two (3%) were discharged. A summary of the specialist input is shown in Table 4.8 overleaf.

The majority of the subjects required a combination of treatments. Nearly half of the subjects required orthodontic treatment for space closure. Of 20 subjects identified for possible treatment with implants, 15 were referred to the Maxillo-facial department for multiple implants with the majority requiring bone augmentation. Adhesive bridges (n=19) and composite build ups (n=17) were often a recommended treatment, in comparison to veneers (n=5), partial dentures (n=6) and overdentures (n=3). Only three subjects required gold onlays on infraoccluded primary second molars. Retained and/or submerged primary teeth were the main indicator for proposed extraction(s).

Table 4.8 Summary of speciality input and treatment.

Specialities	Number of speciality combinations
R	n=6 (8.5%)
P	n=1 (1.4%)
O	n=8 (11.3%)
O+R	n=19 (26.8%)
O+M	n=3 (4.2%)
O+P	n=1 (1.4%)
O+R+M	n=4 (5.6%)
O+R+M+P	n=1 (1.4%)
O+R+P	n=2 (2.8%)
M+P	n=2 (2.8%)
M+R+P	n=1 (1.4%)
M+R	n=1 (1.4%)
OS+O	n=4 (5.6%)
OS+O+M	n=1 (1.4%)
OS+R+O	n=9 (12.7%)
OS+R+O+M	n=2 (2.8%)
OS+O+P+M	n=1 (1.4%)
Monitor	n=3 (4.2%)
Discharged	n=2 (2.8%)
Total	71

R- Restorative/ Paediatric dentistry

P- Prosthodontics

O- Orthodontics

M- Maxillo-facial

OS- Oral surgery

4.2 Part 2: Evaluation of the self-esteem of adolescents with hypodontia.

4.2.1 Sample.

4.2.1.1 The hypodontia study group.

Of the 71 subjects on the hypodontia database, 39 were within the required age range of 13-18 years and subsequently, subject to the other selection criteria, were potential recruits. Twenty-eight subjects were recruited.

Of the 11 not recruited:

- nine were examined for the hypodontia database prior to ethical approval being granted for the controlled study.
- two were undergoing treatment.

4.2.1.2 The control group.

From a potential of 52 subjects aged 13-18 years old, 30 subjects were invited and agreed to participate in the study as part of the control group.

Of the 22 not recruited:

- 13 failed to attend at the Community Dental Clinic, Clydebank.
- five were undergoing orthodontic treatment.
- four subjects were diagnosed with hypodontia.

4.2.1.3 Sample descriptive statistics.

A summary of the descriptive statistics is shown in Table 4.9 below and Table 4.10 overleaf.

4.2.1.4 Age.

The age of those who participated in the hypodontia study group ranged from 13 to 17 years with a mean age of 14.5 years.

The age of those who participated in the control group ranged from 13 to 18 years with a mean age of 14 years.

Table 4.9 The descriptive statistics in terms of age within the hypodontia and control groups.

	Hypodontia Study Group (n=28)	**Control Group (n=30)**
Age (years):		
The mean age	14.5 years	14 years
The modal age	15 years	13 years
The maximum age	17 years	18 years
The minimum age	13 years	13 years
Q1 First Quartile	13 years	13 years
Median	15 years	14 years
Q3 Third Quartile	15 years	15 years

The age of the groups was not normally distributed, given the very small range of ages available. There was no significant difference between the two groups [Mann-Whitney p=0.080].

4.2.1.5 Gender and race.

Of the 28 in the hypodontia study group, females accounted for 19 and males for 9. This represents 68% and 32% respectively of the total sample.

Of the 30 in the control group, females accounted for 16 and males for 14. This represents 53% and 47% respectively of the total sample.

All the subjects in the hypodontia study group and the control group were Caucasian.

Table 4.10 The descriptive statistics in terms of gender within the hypodontia and control groups.

Gender	**Hypodontia Study Group (n=28)**	**Control Group (n=30)**
Males	n=9 (32%)	n=14 (47%)
Females	n=19 (68%)	n=16 (53%)

There was no significant difference in gender distribution between the two groups [chi-squared test, p-value=0.257].

4.2.1.6 Deprivation.

For the hypodontia study group, six participants were categorised as DEPCAT 1 and 2, the most affluent areas, whereas in DEPCAT 7, the most socially deprived area there were five participants. The most common DEPCAT category was 5.

For the control group, one participant was categorised as DEPCAT 1 and 2, the most affluent areas, and in DEPCAT 7, the most socially deprived areas there was also one participant. The most common DEPCAT category was 6.

Further detail is shown in Table 4.11 overleaf.

Table 4.11 Distribution of participants in respect of DEPCAT categories.

Carstairs Deprivation Category Index (DEPCAT)	Hypodontia Study Group (n=28)	Control Group (n=30)
1	n=1 (3.6%)	n=1 (3.3%)
2	n=5 (17.8%)	n=0 (0%)
3	n=3 (10.7%)	n=2 (6.7%)
4	n=5 (17.9%)	n=5 (16.7%)
5	n=9 (32.1%)	n=4 (13.3%)
6	n=0 (0%)	n=17 (56.7%)
7	n=5 (17.9%)	n=1 (3.3%)

4.2.2 The dental status of the control group.

A retrospective examination of the complete dental records of the control subjects reported no complaint at any time from the subjects regarding discolouration or appearance of anterior teeth with the exception of those who had an anterior tooth fracture due to trauma. The descriptive statistics of the dental status of the control group attending the Community Dental Clinic for routine dental treatment is shown in Table 4.12 below.

Table 4.12 Descriptive statistics on the dental status of the control group.

Dental status of the control group n=30	Number of subjects
Anterior caries experience	n=8 (26.6%)
Discolouration due to caries	n=2 (6.7%) n=1 (3.3%) 'invalid' GSEQ
Discolouration due to trauma (slight)	*n=1 (3.3%) n=1 (3.3%) 'invalid' GSEQ
Previous anterior trauma (treated) Maxillary n=3 (10%) Mandibular n=1 (3.3%)	n=4 (13.3%) *n=2 (6.6%) 'invalid' GSEQ n=1 (3.3%) 'invalid' GSEQ
Enamel hypoplasia	n=0 (0%)
Fissure sealants only	n=6 (20%)
Posterior conservation and fissure sealants	n=11 (36.7%)
Posterior conservation	n=10 (33.3%)
Extractions	n=4 (13.3%)

* denotes same subject accounted for under maxillary trauma and discolouration.

4.2.3 Presenting complaint of the hypodontia study group.

From the subject's records the presenting complaint of 20 (71%) of the sample of the hypodontia study group was related to appearance of the teeth. Seven (25%) of the sample did not complain about appearance and one (4%) the presenting complaint was not recorded by the examining clinician. Further data, from the Hosey and Bradnock questionnaire, found that out of the sample of 28, 18 (64%) complained specifically about spaces and gaps.

4.2.4 Congenitally missing teeth in the hypodontia study group.

- The total number of congenitally missing teeth, in the 28 subjects was 256.
- The number of congenitally missing teeth (excluding third molars), in this sample was 189. Of these, 48 teeth were missing from the upper anterior region.
- The total number of "not recorded" teeth was 17.
- The maximum number of congenitally missing teeth (excluding third molars) in a subject was 23.
- The minimum number of congenitally missing teeth (excluding third molars) in a subject was one tooth.
- The mean number of missing teeth (excluding third molars) was seven teeth.
- Twenty-one subjects out of the sample were missing one or more upper anterior teeth.

Further details are summarised in Table 4.13 overleaf.

Table 4.13 Congenitally missing teeth in the hypodontia study group.

	Max. third molars	Max. second premolars	Max. canines *	Max. laterals incisors	Mand. canines *	Mand. second premolars	Mand. third molars
Congenitally missing	n=30	n=24	n=14	n=34	n=10	n=31	n=37
Not recorded	n=6	n=0	n=0	n=0	n=1	n=0	n=6

*There were eight subjects in total with missing canines.

4.2.5 The Hosey-Bradnock questionnaire.

Overleaf are the results of the Hosey-Bradnock questionnaire which was issued and completed by all the participants in the hypodontia study group only. The Hosey-Bradnock questionnaire assessed how the participants felt both about their teeth in general and what they perceived to be their treatment need. The results are shown in Tables 4.14 to 4.21. The answers to the open questions:- (1) What would you want dental treatment to do for you? (2) What do you expect from this visit to the Hypodontia Clinic today? (3) What kind of dental treatment do you expect in the future?- are detailed in Appendix 10.

Table 4.14 What is it about your teeth you don't like?

Loose.	n=0 (0%)
Spaces / gaps.	n=13 (46.4%)
Spaces / gaps. Shape of teeth.	n=3 (10.7%)
Shape of teeth. Teeth look uneven.	n=4 (14.3%)
Spaces / gaps. Shape of teeth. Teeth look uneven.	n=1 (3.6%)
Spaces / gaps. Teeth look uneven	n=1 (3.6%)
Teeth look uneven.	n=3 (10.7%)
Shape of teeth.	n=2 (7.1%)
Omitted	n=1 (3.6%)

Table 4.15 How do you feel about how your teeth look?

hate	dislike	not bothered	Like	happy
n=7 (25%)	n=11 (39.3%)	n=9 (32.1%)	n=1 (3.6%)	n=0 (0%)

Eighteen (64%) of the sample "hated" or "disliked" their teeth.

Table 4.16 Do you feel self conscious or uncomfortable when you smile for photographs, at parties or meet new people?

not at all	a little	fairly	a lot	extremely
n=4 (14.3%)	n=11 (39.3%)	n=5 (17.9%)	n=4 (14.3%)	n=4 (14.3%)

Table 4.17 Do you feel embarrassed about your teeth?

not at all	a little	fairly	a lot	extremely
n=6 (21.4%)	n=13 (46.4%)	n=0 (0%)	n=7 (25%)	n=2 (7.1%)

Table 4.18 Do you get teased at school because of your teeth?

not at all	sometimes	fairly often	Often
n=22 (78.6%)	n=6 (21.4%)	n=0 (0%)	n=0 (0%)

Table 4.19 How much does having missing teeth bother you?

not at all	a little	quite a bit	upsets me
n=4 (14.3%)	n=13 (46.4%)	n=9 (32.1%)	n=2 (7.1%)

Table 4.20 What do you think about wearing false teeth (dentures)?

happy to wear one	would wear one but not happy about it	would never wear one
n=3 (10.7%)	n=15 (53.6%)	n=10 (35.7%)

Table 4.21 What do you think about wearing braces?

happy to wear one	would wear one but not happy about it	would never wear one
n=12 (42.8%)	n=13 (46.4%)	n=3 (10.7%)

4.2.6 The Culture-Free Self-Esteem Inventory.

A total of 58 participants completed the Culture-Free Self-Esteem Inventory from May 2004 to May 2005. This was comprised of 28 from the hypodontia study group and 30 from the control group.

4.2.6.1 Global Self-Esteem Quotient.

The self-esteem of both groups, as measured by the Global Self-Esteem Quotient (GSEQ), is shown in Table 4.22.

Table 4.22 Comparison of the GSEQ of the control and hypodontia study groups.

	Controls (n=30)	**Hypodontia (n=28)**
Mean	**108.4 (s.d. 10.88)**	**108.4 (s.d. 12.09)**
Mode	109	122
Range	77-123	82-122

4.2.6.2 Standard Scores for subscales.

These subscales represent the subcategorisation of self-esteem into academic, general, parental/home, social and personal self-esteem. The subscales yield further information regarding the subjects' strengths and weaknesses. These are shown in Table 4.23 overleaf.

Table 4.23 Comparison between both groups for Subscale Scores.

		Controls	Hypodontia
Academic	*Mean*	11.3	11.3
Academic	*Mode*	13	13
Academic	*Range*	5-13	5-13
General	*Mean*	9.9	9.9
General	*Mode*	10	10
General	*Range*	2-13	2-13
Parental/Home	*Mean*	12.4	12.3
Parental/Home	*Mode*	13	13
Parental/Home	*Range*	9-14	6-14
Social	*Mean*	11.5	11.7
Social	*Mode*	13	13
Social	*Range*	3-13	6-13
Personal	*Mean*	11.3	11.1
Personal	*Mode*	14	14
Personal	*Range*	7-14	3-14

4.2.6.3 Standard Scores in relation to descriptive category of self-esteem.

Using the examiners manual, the Standard Scores for subscales were translated into verbal descriptors of self-esteem. These are shown for the hypodontia and control groups in Tables 4.24 and 4.25 respectively.

Table 4.24 The number of hypodontia subjects (n=28) in each self-esteem subcategory.

Self-esteem descriptor	Academic	General	Parental /Home	Social	Personal	GSEQ
Very low	n=0 (0%)	n=1 (3.6%)	n=0 (0%)	n=0 (0%)	n=1 (3.6%)	n=0 (0%)
low	n=1 (3.6%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=1 (3.6%)	n=0 (0%)
Below average	n=2 (7.1%)	n=2 (7.1%)	n=1 (3.6%)	n=2 (7.1%)	n=2 (7.1%)	n=3 (10.7%)
Average	n=10 (35.7%)	n=18 (64.3%)	n=10 (35.7%)	n=13 (46.4%)	n=13 (46.4%)	n=9 (32.1%)
Above average	n=15 (53.6%)	n=7 (25%)	n=17 (60.7%)	n=13 (46.4%)	n=11 (39.3%)	n=13 (46.4%)
High	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=3 (10.7%)
Very high	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)

Table 4.25 The number of control subjects (n=30) in each self-esteem subcategory.

Self-esteem descriptors	Academic	General	Parental/ Home	Social	Personal	GSEQ
Very low	n=0 (0%)	n=1 (3.3%)	n=0 (0%)	n=1 (3.3%)	n=0 (0%)	n=0 (0%)
low	n=1 (3.3%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=1 (3.3%)
Below average	n=3 (10%)	n=2 (6.7%)	n=0 (0%)	n=1 (3.3%)	n=3 (10%)	n=1 (3.3%)
Average	n=11 (36.7%)	n=22 (73.3%)	n=14 (46.7%)	n=17 (56.7%)	n=18 (60%)	n=12 (40%)
Above average	n=15 (50%)	n=5 (16.7%)	n=16 (53.3%)	n=11 (36.7%)	n=9 (30%)	n=14 (46.7%)
High	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=2 (6.7%)
Very high	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)

4.2.6.4 Defensiveness Score.

4.2.6.4.1 Defensiveness Score in relation to GSEQ.

A score of greater than or equal to four, questions the validity of the GSEQ value. Comparison of the Defensiveness Score in the two groups is detailed in Table 4.26. Furthermore, Table 4.27 indicates the number of subjects, by group, whose GSEQ value was questionable.

Table 4.26 Defensiveness Score in the hypodontia and control groups.

	Number of subjects	
Defensiveness Score	**Hypodontia study group (n=28)**	**Control group (n=30)**
0	n=3 (10.7%)	n=1 (3.3%)
1	n=9 (32.1%)	n=9 (30%)
2	n=6 (21.4%)	n=4 (13.3%)
3	n=3 (10.7%)	n=7 (23.3%)
4	n=0 (0%)	n=3 (10%)
5	n=2 (7.1%)	n=4 (13.3%)
6	n=1 (3.6%)	n=0 (0%)
7	n=4 (14.3%)	n=2 (6.7%)
8	n=0 (0%)	n=0 (0%)

Red font denotes the number of subjects with Defensiveness Scores ≥ 4

Table 4.27 The number of subjects with a questionable GSEQ

	Hypodontia study group (n=28)	**Control group(n=30)**
Defensiveness Score ≥4	n=7 (25%)	n=9 (30%)
Self-esteem recorded	n=2 (7.1%) High self-esteem n=3 (10.7%) Above Average self-esteem n=2 (7.1%) Average self-esteem	n=1 (3.3%) High self-esteem n=5 (16.7%) Above average self-esteem n=3 (10%) Average self-esteem
Defensiveness Score <4	n=21 (75%)	n=21 (70%)

4.2.6.4.2 Defensiveness Scores in relation to descriptors of self-esteem.

The Defensiveness Score in relation to the self-esteem descriptor for the hypodontia and the control groups is shown Tables 4.28 and 4.29 respectively.

Table 4.28

Defensiveness Score of the hypodontia study group (n=28) in relation to self-esteem descriptors.

	Defensiveness Score								
Self-esteem	**0**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**
Very low	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)
Low	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)
Below average	n=0 (0%)	n=3 (10.7%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)
Average	n=2 (7.1%)	n=2 (7.1%)	n=2 (7.1%)	n=1 (3.6%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=2 (7.1%)	n=0 (0%)
Above average	n=1 (3.6%)	n=3 (10.7%)	n=4 (14.3%)	n=2 (7.1%)	n=0 (0%)	n=0 (0%)	n=1 (3.6%)	n=2 (7.1%)	n=0 (0%)
High	n=0 (0%)	n=1 (3.6%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=2 (7.1%)	n=0 (0%)	n=0 (0%)	n=0 (0%)
Very high	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)

Red font denotes the number of subjects with Defensiveness Scores ≥ 4 in each of the self-esteem descriptors.

Table 4.29

Defensiveness Score of the control group (n=30) in relation to self-esteem descriptors.

	Defensiveness Score								
Self-esteem	**0**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**
Very low	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)
Low	n=0 (0%)	n=1 (3.3%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)
Below average	n=0 (0%)	n=1 (3.3%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)
Average	n=1 (3.3%)	n=2 (6.7%)	n=2 (6.7%)	n=4 13.3%)	n=1 (3.3%)	n=2 (6.7%)	n=0 (0%)	n=0 (0%)	n=0 (0%)
Above average	n=0 (0%)	n=5 (16.7%)	n=2 6.7%)	n=2 (6.7%)	n=2 (6.7%)	n=1 (3.3%)	n=0 (0%)	n=2 (6.7%)	n=0 (0%)
High	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=1 (3.3%)	n=0 (0%)	n=1 (3.3%)	n=0 (0%)	n=0 (0%)	n=0 (0%)
Very high	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)	n=0 (0%)

Red font denotes the number of subjects with Defensiveness Scores ≥ 4 in each of the self-esteem descriptors.

4.2.6.5 Comparison of GSEQ values in 'truthful' subjects between hypodontia and control groups.

There were 21 'truthful' subjects in both the hypodontia and control groups. In respect to the control group, there were 11 females and 10 males and their mean age was 14 years (13 to 18). In respect to the hypodontia study group, there were 16 females and five males and their mean age was 14.6 years (13 to 17). Table 4.30 overleaf shows the number of congenitally missing teeth in relationship to descriptors of self-esteem of the 21 'truthful' subjects in the hypodontia group.

Table 4.30 The number of congenitally missing teeth in relationship to descriptors of self-esteem of the 'truthful' subjects in the hypodontia group.

	Descriptor of self-esteem for 'truthful' subjects in the hypodontia group (n=21)			
	High self-esteem (n=1subject) (4.7%)	Above average self-esteem (n=11 subjects) (52.4%)	Average self-esteem (n=6 subjects) (28.6%)	Below average self-esteem (n=3 subjects) (14.3%)
Number of congenitally missing teeth	n=3	n=16	n=12	n=7
		n=13	n =11	n=5
		n=12	n=10	n=4
		n=14	n=8	
		n=12	n=2	
		n=10	n=1	
		n=8		
		n=6		
		n=4		
		n=3		
		n=1		
Range		1-16	1-12	4-7
Average	n=3	n=9	n= 7	n=5

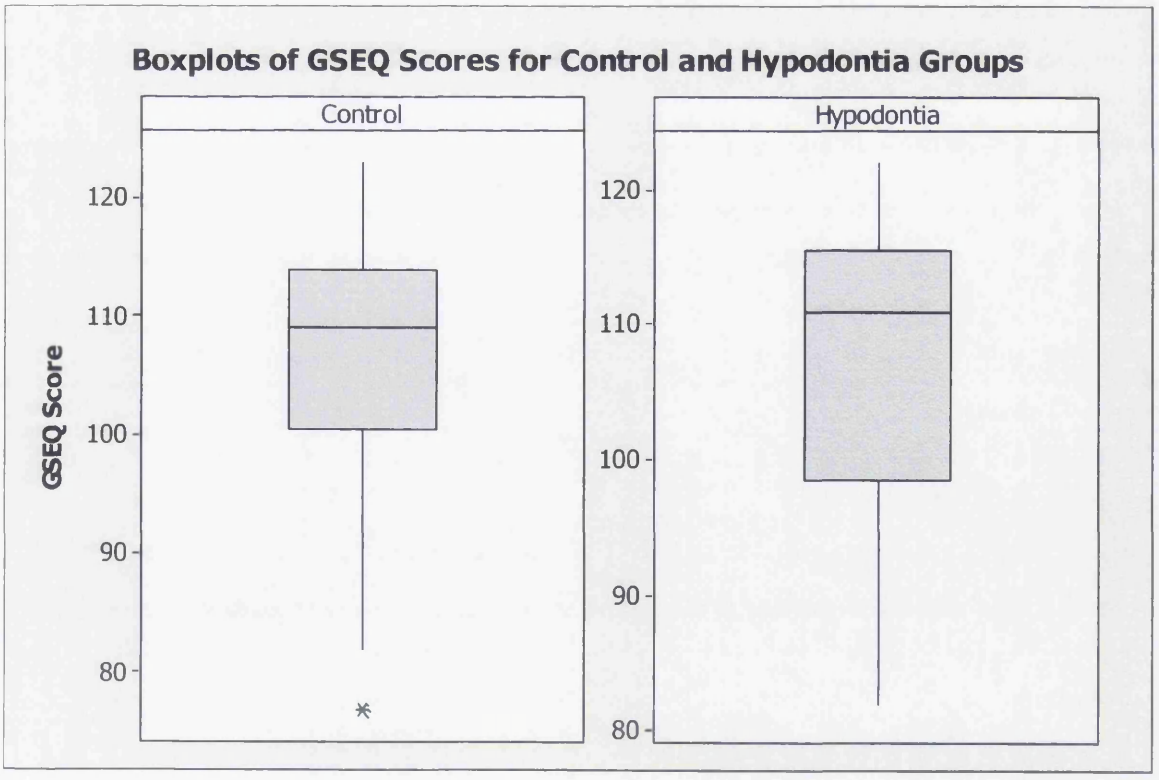
The descriptive statistics are summarised below in respect of the GSEQ values for the 'truthful' subjects in Table 4.31 and Figure 4.1 using medians and ranges, given that the data was not normally distributed. There was no significant difference between the two groups [Mann-Whitney p=0.8].

Table 4.31 The GSEQ data summarised in medians and ranges.

	Minimum	Q 1	Median	Q3	Maximum
Control group	77	100.50	109.00	114.00	123.00
Hypodontia Group	82	98.50	111.00	115.50	122.00

Q1 denotes first quartile. Q3 denotes third quartile

Figure 4.1 Boxplots of the GSEQ data for the control and hypodontia groups.



4.2.6.6. Feelings and descriptors of self-esteem in relation to congenitally missing permanent anterior teeth.

Fifteen of the 'truthful' hypodontia subjects had upper anterior teeth missing. Their responses concerning how they felt about their teeth are shown in Table 4.32 below and the relationship of the number of missing anterior permanent teeth to descriptors of self-esteem is shown in Table 4.33 overleaf.

Table 4.32 The reported feelings of truthful hypodontia subjects about how they feel about having missing upper anterior teeth.

	How do you feel about how you teeth look?				
(n=15)	Hate	Dislike	Not bothered	Like	Happy
Number of subjects	n=4 (26.7%)	n=7 (46.7%)	n=4 (26.7%)	n=0 (0%)	n=0 (0%)

Table 4.33 The relationship of the number of missing anterior permanent teeth to descriptors of self-esteem.

Number of permanent anterior teeth missing	Number of 'truthful' subjects (n=15)	Descriptor of self-esteem
n=4	n=3 (20%)	Above average self-esteem n=3 (20%)
n=2	n=7 (46.7%)	Above average self-esteem n=1 (6.7%) Average self-esteem n=5 (33.3%) Below average self-esteem n=1 (6.6%)
n=1	n=5 (33.3%)	Above average self-esteem n=3 (20%) Average self-esteem n=2 (13.3%)

4.2.6.6 Adequacy of sample size.

For the benefit of future study, a statistical power calculation, using the standard deviation based on the 'truthful' control group GSEQ scores (s.d. 11.62), to examine for a difference between the two groups, where a difference of ten was deemed to be of clinical relevance (since this related to the value required to effect an alteration in self-esteem descriptor) confirmed that using a two-sample t-test and five percent significance level and 80% power, 23 subjects would be required in each group. Maintaining the same parameters but increasing the power to 90%, 30 subjects would be required in each group. If the standard deviation from the combined groups was used (s.d. 11.83), the required sample sizes would change little. Furthermore, if the standard deviation from the "truthful" hypodontia group (s.d. 12.33) was used the required sample sizes would be 25 and 30 subjects per

group, for 80% and 90% power respectively. Table 4.34 shows the GSEQ mean and standard deviation values.

Table 4.34 Mean and standard deviation of the "truthful" GSEQ values.

	Mean	Standard Deviation (s.d.)
Control group	105.67	11.62
Hypodontia Group	106.00	12.33
Combined	105.83	11.83

4.3 Summary of Results.

4.3.1 Part 1: The profile of subjects at the Hypodontia Clinic:

1. The most frequent age for referral to the Hypodontia Clinic was 12 years and there was a female to male ratio of 2:1. The social deprivation of those referred matched that of the catchment area.
2. A positive family history was reported by 42% of hypodontia subjects.
3. Sixty eight percent of hypodontia subjects complained about appearance of their teeth.
4. The majority of hypodontia subjects presented had Class I incisal relationships with normal or decreased overjets.
5. The mean number of missing teeth (excluding third molars), was six teeth with a range of one to 23 teeth. The most common reported missing tooth was the maxillary lateral incisor.
6. Fifty four percent of subjects had six or more congenitally missing teeth.
7. Eighteen subjects had missing canines.
8. Three subjects had phenotypes that corresponded to specific gene types involved in hypodontia.
9. Thirty-four subjects showed phenotypes of a severe extended pattern of hypodontia.
10. The majority of the subjects subsequently underwent combined orthodontic and restorative treatment, mainly in Orthodontic and Paediatric dentistry.
11. The favoured treatment options were that of space closure, composite build ups, adhesive bridges and implants.

4.3.2 Part 2: Self-esteem of adolescents with hypodontia:

1. Adolescents with hypodontia did not have a lowered self-esteem compared to non-hypodontia controls.
2. In the hypodontia study group, 71% complained about the appearance of their teeth with the vast majority (64%) specifying spaces and gaps.
3. Twenty-one of the hypodontia subjects were missing one or more upper anterior teeth.
4. Eighteen (64%) of the hypodontia subjects "hated" or "disliked" their teeth. Over three quarters of them reported that they were self-conscious and embarrassed about their teeth. However, 22 (79%) reported that they did not get teased at school. Eighty-six per cent reported that having missing teeth bothered them.
5. Only three subjects (11%) were happy to wear a denture, 53% would wear a denture but would not be happy about it and 36% would never wear one. In respect to orthodontic appliances, 43% were happy to wear one and a further 46% would wear one but would not be happy about it and 11% would never wear one.

Chapter 5 Discussion.

5.1 Part 1: The profile of subjects at the Hypodontia Clinic.

5.1.1 Participation.

There was a high level of participation in compiling the hypodontia database. Indeed, all who were invited participated.

5.1.2 Age.

The results of the present study, for the modal age of the sample, presenting at the Hypodontia Clinic were similar to that reported by Hobkirk et al (1994). The aforementioned authors reported that the majority of subjects in their sample were between eight and thirteen years of age at their first attendance at the Hypodontia Clinic.

Hobkirk et al (1994) reported that over 55% of their subjects were over 12 years old at the time of presentation at the Hypodontia Clinic. They suggested that delayed or late referrals resulted from poor recognition and a lack of knowledge about hypodontia by the referring dentists, together with low levels of demand by subjects and/or difficulty in finding an appropriate clinic to which to refer the subject. In the present study, just over 60% of those presenting at the Hypodontia Clinic over 12 years of age.

5.1.3 Gender, race and social deprivation.

With respect to the gender split of the subjects in the database, this was anticipated, in that more females than males were recorded. The 1 : 2,

male : female ratio is in accordance with the generally accepted ratio of 2 : 3 (Egermark-Eriksson and Lind 1971). It was anticipated that this sample would be predominately Caucasian as this reflected the population of the referral area. The fact that the majority of the subjects came from socially deprived areas is also in accordance with the population in the referral area (McLoone 1994).

5.1.4 Family history.

Even though almost half reported a positive family history of hypodontia, it is suspected that this may be an underestimation, especially with evidence of the genetic nature of the condition (Grahnén 1956). Despite this reserve, our result for a positive family is slightly higher than that reported from a similar study of 59 subjects with hypodontia attending the Hypodontia Clinic at Newcastle Dental Hospital (Chung et al 2000). In some cases, family histories may be recorded incorrectly, negative, as a result of lack of knowledge of family history because of deaths of older family members, family break-ups, adoption or simply a lack of knowledge or interest in the condition.

5.1.5 Presenting complaints.

The predominant complaint of the subjects was that of poor appearance. The present study participants highlighted the greatest concern being that of spaces, especially within the anterior region. Again, these results are accordance with the study by Hobkirk et al (1994). However, Chung et al (2000) reported that not only was poor appearance a significant presenting complaint but also that of lack of function. However, in the present study, lack of function was rarely reported.

Indeed, Tulloch et al (1984) reported that dental appearance rather than health and function is the main motivating factor in seeking orthodontic treatment.

Furthermore, previous researchers (Shaw et al 1979, Shaw 1981b, Kilpelainen et al 1993) have confirmed that females are more likely to be concerned about the appearance of their teeth and actively seek orthodontic treatment.

5.1.6. Incisal relationship.

Despite only two-thirds of the incisal relationships of the subjects being recorded, the number with Class II Division II incisal relationship was lower than expected. A three fold increase in congenital tooth anomalies compared to the normal population has been reported associated with this incisal relationship (Basdra et al 2001). In the present study, Class I incisal relationships were the most prevalent, which concurs with the study of Rose (1966).

An increased overjet has been reported as being a high indicator in those seeking orthodontic treatment because of concern about appearance, teasing and self-esteem (Horrowitz et al 1971, Shaw et al 1980, Shaw 1981b, Tulloch et al 1984, Kilpelainen et al 1993). Even though most of the subjects in the present study did not have an increased overjet they were still sufficiently concerned about their missing teeth to actively seek treatment by referral to the hypodontia clinic by their primary care practitioner.

5.1.7 Skeletal base.

The skeletal base in the present study was judged visually. Therefore, the results of the skeletal base analysis cannot be compared against studies using lateral cephalograms (Chung et al 2000). The aforementioned authors reported that severe hypodontia showed tendencies towards Class III skeletal relationship. Despite nearly two thirds of the subjects in the present study showing six or more teeth congenitally missing, a normal skeletal relationship was the most prevalent of those recorded.

5.1.8 Congenitally missing teeth.

In this study, subjects and their parents/guardians were questioned regarding the subjects past dental history, with specific reference to previous extractions of permanent teeth, so that extracted teeth were not charted as congenitally missing in error.

5.1.8.1 Most common missing tooth.

In this study, the most common missing tooth, excluding third molars, was the maxillary lateral incisor followed closely by the mandibular second premolar. This was an anticipated result as in accordance with the other prevalence studies in Caucasians where the third molar has been excluded (Dolder 1937, Brekhus et al 1994, Grahnén 1956, Muller 1970).

5.1.8.2 Multiple missing teeth.

The results of the present study also reflect the findings of Rolling (1980) and Grahnén (1956) in that hypodontia in the premolar region and lateral incisor region predominate.

It should be born in mind that the Hypodontia Clinic at Glasgow Dental Hospital and School, is within the tertiary clinical pathway. Mild hypodontia can be adequately treated by practitioners in the primary care setting. Therefore, the subjects in the sample of this study tended to present with more severe hypodontia. Furthermore, the number of peg shaped laterals reported was lower than anticipated, but this reflected the findings of Lai and Seow (1989) in that, in those with multiple missing teeth, the incidence of peg-shaped laterals was reported to fall.

5.1.8.3 Congenitally missing canines.

The literature suggests that canines are one of the "stable" teeth in the dentition (Dalberg 1945). However, in the present study, there was an unexpected and unexplained preponderance of missing canines which accounted for over a quarter of the sample. The majority of these missing canines were from the maxilla. It has been reported that, although missing canines are rare, when this does occur, it is in cases of severe hypodontia (Brekhus et al 1944, Hobkirk and Brook 1980). Indeed, Hobkirk et al (1994) also reported a notable number of maxillary canines as well as second molars in their sample of 451 subjects with hypodontia. The aforementioned authors presumed this was related to the number of severely affected subjects. Few prevalence studies mention hypodontia of canines, but

Cameron and Sampson (1996) reported a prevalence of 0.4% for maxillary canines. In the present study, those subjects who had maxillary canines missing, often the adjacent lateral incisor was missing as well. This might be a genetic effect operating in an anterior orofacial field as with other canine anomalies (Peck et al 1996b).

Of the sample of hypodontia subjects (n=71), only one set of siblings presented at the hypodontia clinic and both presented with congenitally missing canines. Investigation of their pedigree in depth may be of interest in respect of the missing canines. It also highlights the important role that the hypodontia database can play in the tracking of hypodontia in families as it is appears unusual for siblings to present at the same time at the Hypodontia Clinic.

5.1.8.4 Patterns of missing teeth.

The literature has reported hypodontia in terms of the number of teeth missing and the most common missing tooth. However, in this present study the patterns of missing teeth have been recorded in an attempt to correlate this to the known genetic markers in humans (Vastardis et al 1996, Arte et al 1996, Stockton et al 2000, Nieminen et al 2001).

It was very difficult to extrapolate set patterns as there were so many variations in the patterns of missing teeth. Very few subjects fell into the specific phenotypes expressed by Msx 1 and Pax 9 according to the literature (Nieminen et al 1995, Vastardis et al 1996, Arte et al 1996, Stockton et al 2000, Nieminen et al 2001). The results in this study show that the majority of subjects fall into a range of

phenotypes, possibly corresponding to mutations or differences in expression of the Msx 1 and Pax 9 genes. From the results in this sample, it seems that these genes rarely have an effect solely but appear to act in a combination, to effect these phenotypes (Vieira et al 2004, Mostowska et al 2003a). The results show how complex the trait of hypodontia is, with the involvement of genes and their likely interaction with one another. Another gene, TGFA may be involved in these subjects. TGFA is thought to be involved in hypodontia of incisors (Vieira et al 2004). This implication of TGFA in familial hypodontia has only been documented by the aforementioned authors. There is therefore, a possibility that, a combination of TGFA with all the aforementioned genes may result in the phenotypes in our sample. The high involvement of missing maxillary lateral incisors in the patterns was anticipated, in light of the prevalence studies.

In the present study, only one subject had been diagnosed with having a syndrome. However, the possibility that there are undiagnosed syndromes in this population should be recognised. Chung et al (2000) reported that in their sample of 59 subjects with hypodontia, seven per cent had an associated syndrome. Therefore, it is reasonable to suspect that within this sample there are undiagnosed syndromic subjects, as over half of the sample presented with six or more teeth missing (Schalk-van der Weide 1994). As such, the most likely gene is that of anhidrotic ectodermal dysplasia (EDA) (Kere et al 1996). Furthermore, in those with less severe hypodontia there is the possibility of female carriers of ectodermal dysplasia (McLaughlin 1991, Nunn et al 2003). Therefore, based on the results of this study, the involvement of a geneticist in this multidisciplinary

clinic is to be recommended. Only further genetic analysis of these subjects will confirm or eliminate these hypotheses.

5.1.9 Associated anomalies found with hypodontia.

It was thought that the number of subjects with anomalies associated with hypodontia would have been greater than reported in light of the literature (Peck et al 1993, 1994, 1996a and b, 1998, Lai and Seow 1989, Baccetti 1998a and b).

5.1.10 The assessment form.

In the present study, there were several examiners involved. In studies involving more than one examiner, the "human factor" must be taken into account for possible omissions (Brekhus et al 1944). It is accepted that there are difficulties in charting teeth accurately in subjects with hypodontia (Niswander and Sujaku 1963).

In the present study, the clinical assessment form was an adapted orthodontic assessment form. It transpired not to be "user friendly" to the examining clinicians, so simple changes were made and resulted in an improved assessment with more accurate chartings. Despite these changes there was about a third of the data omitted by the examining dentists on the assessment form in respect of the incisal relationship and size of the overjet, approximately a tenth missing in respect of the skeletal pattern and approximately a quarter missing in respect to spacing/crowding/alignment of the teeth in the dental arches. These clinical observations in most cases will have been recorded in the subject's records but

were not recorded in this assessment sheet. These omissions are due to the "human factor" and the assessment form should be reviewed again in order to improve the overall accuracy. This emphasises the importance of an assessment form fit for purpose.

5.1.11 Treatment options.

Treatment that was planned often related to the management of missing anterior teeth. This involved space closure or spacing opening in order to provide symmetry. Orthodontic treatment was frequently provided in conjunction with restorative dentistry in the form of provision of adhesive bridges or camouflaging canines to mimic missing laterals. Maxillofacial surgery was planned where there was a skeletal deformity present, requiring correction with multiple implants and often bone augumentation. The provision of a partial denture was rare, but was advocated either because of very severe hypodontia or, more usually, was an intermediatory treatment prior to a more definitive treatment plan. The overdenture was proposed either as an "intermediate" treatment to improve aesthetics or to prevent overeruption usually of the lower canines. As in some cases, implants were planned for the future once the subject had ceased growing. An intermediatory treatment plan helped to progress towards this preferred treatment of implants (Jepson et al 2003). Most subjects wished to avoid a denture if at all possible, but were more willing to wear an orthodontic appliance. This is in keeping with other studies (Hobkirk et al 1995, Jepson et al 2003, Meechan et al 2003, Nunn et al 2003).

5.2 Part 2: A prospective controlled study to evaluate the self-esteem of a sample of adolescents with hypodontia

5.2.1 Participation in the study.

There was also a high level of participation in the self-esteem study. Indeed no-one declined to participate in either the study group or the controls. The controls were easier to recruit than expected probably because most were long-term patients of the author, Mrs Rosemary Broad. The controls compared well with the normative sample and gave a good representation within our population (Battle 2002). In fact, Battle (2002) recommended a control sample within the population in which a study group would be analysed.

5.2.2 Sample size.

In the present study, the sample size is comparable to other studies using the Culture-Free Self-Esteem Inventory (Cornman 1993, Bolden and Williams 1995, Iniewicz 2005). Furthermore, the sample size of the present study has been endorsed by the power calculations of the statistician.

5.2.3 The holistic view.

The two questionnaires used, developed an overall picture. The Culture-Free Self-Esteem Inventory recorded the level of self-esteem, but it did not report on "why" a low self-esteem or a high self-esteem had been recorded (Battle 2002). The Hosey-Bradnock questionnaire gave an insight on the "why" question.

5.2.4 Descriptive comparison between the control group and the study group.

5.2.4.1 Age.

The hypodontia study group and control group were well matched for age and this was better than was expected. This was a result of a high attendance rate of adolescents attending the Community Dental Clinic at Clydebank for routine dental treatment. With the similar mean age in both groups, differences in maturity can be disregarded. The age of the subjects in this study was also comparable to other studies of hypodontia subjects (Hobkirk et al 1994, Chung et al 2000).

5.2.4.2 Gender.

The gender ratio for the hypodontia study group was as expected and matched that reported in the database. The gender mix in the controls reflected normal gender distribution. As a result, there were more males in the control group compared to the study group. Recruitment of more female controls would have eliminated this limitation of the study. Nonetheless, the Culture-Free Self-Esteem Inventory has been validated by gender, race, residence, age and educational status.

5.2.4.3 Race.

All the subjects in both the hypodontia and control group were Caucassian. Therefore, comments in respect of race differences cannot be made. However, the Culture-Free Self-Esteem Inventory, as previously mentioned, has been validated in respect of race. The author, Battle (2002) reported that no measure is completely culture-free and that "culture fair" is a more accurate representation of

this measure. Despite this, Battle (2002) have retained "culture-free" within the name of the inventory for historical purposes.

5.2.4.4 Social deprivation.

It was anticipated that for the controls the majority would score DEPCAT 5 and/or 6, as the controls were recruited from the one catchment area, namely Clydebank, in which DEPCAT scores of 5 and 6 predominate (McLoone 1994). The hypodontia group were recruited from throughout Glasgow, which in itself has many socially deprived areas.

Half of the hypodontia group came from DEPCAT 5, 6 and 7 compared to almost three quarters of the controls. Therefore, there is a bias of the control group towards the higher DEPCAT scores and thus from the more socially deprived areas. Burden and Pine (1995) however reported that peer group influences were more significant than social class or gender in the uptake of orthodontic treatment. The results regarding self-esteem appear not to have been influenced by the DEPCAT score. Ideally, to make a direct comparison regarding DEPCAT scores this would have involved other clinics across Glasgow and recruiting more controls to match the DEPCAT scores.

5.2.5 Evaluation of the self-esteem of adolescents with hypodontia.

The adolescents with hypodontia did not have a lowered self-esteem. It was anticipated that the self-esteem of those with hypodontia would be lowered and, furthermore, would be lowest in those with the most teeth missing (Duggal and

Ogden 1990). Indeed, even the subcatergorisation of the subscales indicated no difference, even in the personal subscale. The number of missing permanent anterior teeth did not indicate a difference. In future studies, the measurement of the actual space/gap present opposed to the actual number of missing permanent anterior teeth maybe a more useful measurement, as this would have a greater impact on anterior aesthetics. This finding, is at odds with the literature that reports that self-image and the importance of appearance, especially facial aesthetics play an integral part in self-esteem (Shaw 1981b, Kilpelainen et al 1993, Hobkirk and Jones 1994, Kapp-Simon and McGuire 1997). Interestingly, a study using another self-esteem inventory, Piers-Harris, involving subjects with cleft lip and palate found similarly surprising results, in that most of the subjects recorded self-esteem of average or above average (Leonard et al 1991). Kapp-Simon et al (1992) reported that self-concept scores may not be the best indicators of overall psychological functioning and suggested that a validated measure of adjustment would be more appropriate. A similar study to the present study, using a validated measure of adjustment would be interesting.

The attitude of parents, the manner in which the condition has been explained to both the child and parent, and the referral by the primary care practitioner may all have influenced the self-esteem (Brantley and Clifford 1979a, Leonard et al 1991). The literature has reported that self-esteem is comprised of (1) attitudes towards oneself (2) knowing one's attributes and also (3) accepting one's limitations (Battle 2002). The subjects in this study have quite clearly stated their dissatisfaction with their appearance, have come to terms with the fact there is a problem and have, with the help and support of parents, actively sought treatment

to resolve the problem (Kapp 1979, Leonard et al 1991, Dekovic and Meeus 1997). There is also the possibility that just knowing one is going to receive treatment may boost self-esteem (Albino et al 1994). It can be assumed that these subjects and their parents regard teeth, and their appearance, as important, otherwise, they would not have attended. Therefore, they, and their parents have not only coped but also have had the ability to access treatment centres.

Many of the subjects reported feeling self-conscious and/or embarrassed about their teeth and this may account for some subjects appearing reticent and reluctant to smile or show their teeth at their assessment appointment. On the other hand, in some subjects this behaviour, could be due to attending an unfamiliar environment and the uncertainty of what was going to happen at the appointment. This reticence could be possibly regarded as a defence coping mechanism (Kapp-Simon et al 1992, Leonard et al 1991).

5.2.6 The Defensiveness Score.

The "lie detector" within the Culture-Free Self-Esteem Inventory indicates those individuals whose self-esteem score is invalid. It was interesting that the self-esteem differed little when the invalid self-esteem results were omitted. This finding probably highlights that not wishing to divulge your true feelings is a normal trait in the population.

Culture-Free Self-Esteem Inventory manual reports that those recording a low self-esteem have, low self-esteem, as a result of a low self-opinion of themselves. Whereas, those recording high self-esteem may have, high self-esteem as a result

of maturity, a high self opinion of themselves or alternatively are masking a low self-esteem with a high self-esteem (Battle 2002). Thus, in the present study, there is the possibility that a subject with a low self-esteem could be masking it, by giving themselves a more favourable self-esteem through "lying". However, this is not borne out in the findings. Other self-esteem questionnaires do not have "a lie detector". Therefore, the findings of the present study, where only valid responses were included in the analysis make this more robust.

5.2.7 Completion of the Culture-Free Self-Esteem Inventory.

There was no missing data as the form was easy to use and the subjects were supported by the researcher. The Culture-Free Self-Esteem Inventory was age specific, and the majority of the subjects found little problem understanding the questions. Interestingly, there was only one word, "confide", of which a few subjects in the control group asked for the meaning. There was no request from any of the hypodontia subjects for this or any other word.

5.2.8 Hosey-Bradnock questionnaire.

5.2.8.1 Appearance.

The majority of subjects were unhappy with the appearance of their teeth, specifically highlighting spaces and gaps. This was reported in previous studies (Tulloch et al 1984, Hobkirk et al 1994). This finding can be explained by the majority of these subjects having missing maxillary anterior teeth. Again, with the unexpected number of missing maxillary canines together with lateral incisors, resulting in wide anterior spacing, these complaints relating to appearance were

justifiable. Furthermore, some subjects raised concerns of the spaces and gaps that would appear on the exfoliation of primary teeth, especially retained primary canines. Therefore, it is understandable that seeking treatment from the subject's perspective is driven by aesthetics rather than function (Tulloch et al 1984). The influences of the media (Peck and Peck 1970, Shaw 1981c) and the influences of peers also have an effect on the perception of an attractive appearance (Burden and Pine 1995).

The Hosey-Bradnock questionnaire reveals the depths of feelings these subjects have towards the appearance of their teeth. As expected, the depth of feeling towards the subject's appearance of their teeth is principally governed by the spacing caused by the missing teeth and the location. It is of no surprise that the majority of those with maxillary anterior teeth missing elicited feelings of "hate" or "dislike". However, there is an assumption of subjects not liking their teeth in first question of the Hosey-Bradnock questionnaire. One subject highlighted this by omitting the question. Therefore, modification of the questionnaire to include other questions or leaving a blank space for the subject to complete themselves, might be recommended for any future study.

5.2.8.2 Self-consciousness and embarrassment.

The subjects feelings of self-consciousness and embarrassment towards their teeth really mirrors their feelings towards the appearance of their teeth and again reflects on the number with upper anterior teeth missing. Although, many reported being only "a little embarrassed" about their teeth it is suspected this might be an underestimation and they are hurting more than they express.

5.2.8.3 Teasing.

It was gratifying that teasing was not apparent at school. The literature reports that teasing is related to those with Class II malocclusions with increased overjet (Shaw et al 1980, Kilpelainen et al 1993) and to those with a craniofacial abnormality (Turner et al 1997). The lack of reported teasing in the present study may be accounted for that the majority of the subjects tended to have Class I malocclusion with a normal or decreased overjet. There is a possibility that this lack of teasing in childhood prevents a lowered self-esteem.

5.2.8.4 Attitude to dentures.

The subjects' attitude to wearing a denture was surprising, with only slightly more than a third reporting they "would never wear one" and over half reporting they "would wear one but not happy about it". It was anticipated that the number of those selecting "never to wear" a denture would be higher. These results possibly indicate a reflection to the degree that, these subjects would put up with wearing a denture, although not happy about it to improve aesthetics. Clearly bridges are considered more "socially acceptable" than dentures when you are young.

5.2.8.5 The open questions.

The last three questions allowed the subjects to express what they wanted with regard to their teeth and what they expected from this appointment and, in the future. Their answers were very frank and to the point. The majority were seeking what they considered "normal" looking teeth. These hand written comments in

immature hand writing said more than the questionnaire data, as this is individual children writing from the heart. To sum up: it was a "cry for help".

Some participants were very knowledgeable about what treatment was available and had heard or read about bridges and implants from the media. Others were seeking information from this appointment and in most cases, enthusiastically agreed to embark on often complicated treatment plans. A concern for clinicians is that subjects may have unrealistic expectations of the treatment outcome. A study by Kiayk et al (1982), highlighted this problem after orthognathic surgery. With some subjects, their expectations of surgery were unrealistic, in that, it would change their lives. As a result of this perceived failure, their self-esteem plummeted about nine months after surgery despite having been normal both prior to surgery and at a four month assessment post surgery. Subjects post treatment may report dissatisfaction with other facial aspects which previously had gone unnoticed prior to treatment of the initial complaint/problem (Kiayk et al 1982). Therefore, clinicians must be aware of this, and carefully explain to the subjects that whilst they will feel happier about their appearance, this surgery alone will not change their lives (Richman 1983).

5.2.9 Completion of the Hosey-Bradnock questionnaire.

There was no missing data from the Hosey-Bradnock questionnaire, except from the intentional omission of the response of question one from one subject, previously mentioned under *6.2.7.1 Appearance.*

5.2.10 Bias within the hypodontia study group.

The hypodontia study group may be considered a bias sample as they were being assessed for treatment for the condition. To ascertain this, a population survey would have to be completed which involved a clinical and radiographic examination. This would have practical and ethical difficulties.

Furthermore, within the Glasgow Dental Hospital subjects with hypodontia are also examined and treated in the the Dento-facial clinic and also the normal orthodontic clinic, when a restorative opinion is often sought directly. Therefore, there is bias of the results as the results are from the hypodontia clinic only.

5.2.11 Bias within the control group.

The majority of the control subjects were long-term patients of the researcher, Mrs Rosemary Broad, and as a result this may have led to bias in their results. However, despite this possible bias, their scores matched the normative sample for the inventory and this is the only study that has included a control group.

The control group represented a typical group of subjects attending a Community Dental Clinic for routine dental treatment and therefore represented a normative sample of the population for the 13-18 age group. In an ideal situation the selection criteria for the controls would only have included those with absence of previous anterior carious experience, absence of a history of trauma, absence of enamel hypoplasia, not undergoing orthodontic treatment and without hypodontia. These criteria would have eliminated almost half of this control sample and there were no other potential recruits available at this clinic. To involve other clinics in

the study would have been logistically difficult. As a result, there may be some bias regarding inclusion of those who had enamel hypoplasia, discolouration because of caries or non-vitality and/or previous trauma to anterior teeth. Therefore, those so affected, could be considered inappropriate controls, as their self-esteem may have been affected by the appearance of their teeth. Indeed, of those subjects with discolouration and trauma (n=6), two thirds had an 'invalid' GSEQ value and were therefore excluded from the later statistical analysis of the comparison of the GSEQ values of 'truthful' subjects in the hypodontia and control groups.

5.2.12 Further research.

Often a research study raises more questions than answers on completion. It is gratifying that these adolescents with hypodontia do not appear to have a lowered self-esteem. However, the issue of an unrealistic expectation of treatment outcomes is real. Review of these subjects post-treatment using the Culture-Free Self-Esteem Inventory with a "treatment satisfaction" questionnaire would be able to assess this. It would be interesting to compare the post-treatment self-esteem of subjects with hypodontia to the findings of the post treatment self-esteem of mild to moderate malocclusions by Albino et al (1994). Data from this would benefit future subjects as a result of a quality assurance audit. It would possibly highlight problems from the subjects' perspective not obvious to the clinicians.

Another future study would be a study of the self-esteem and feelings of the parents and/or older siblings who were also affected by hypodontia. Again, there

maybe data from these individuals which is of importance with regard to the future ongoing management of subjects with this condition.

In this present study, the patterns of congenitally missing teeth have been explored in context with the molecular biology literature of the genes found to cause hypodontia. An exciting future study would be to complete genetic analysis on these subjects with distinct patterns of missing teeth. Furthermore, through further research the question of the unexplained and unexpected preponderance of congenitally missing canines may be answered. Indeed, the dental profession are in a unique situation to have the ability to diagnosis. In the light of a suspicion that a good number of our sample may have undiagnosed syndromes, it may be considered a moral and professional duty, to instigate genetic analysis for these subjects. This brings with it the consideration of the importance of genetic counselling.

Chapter 6 Conclusion.

6.1 The conclusions.

The subjects attending the Hypodontia Clinic at Glasgow Dental Hospital and School represented a typical sample of hypodontia subjects, except for the preponderance of missing maxillary canines.

Self-esteem was not lowered in adolescents with hypodontia.

6.2 The null hypothesis.

"There is no difference in self-esteem of subjects with hypodontia compared to non-hypodontia controls"

The null hypothesis is accepted

Appendices.

Appendix 1.

Ethical approval for the Hypodontia Clinic database.

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Dr Marie Therese Hosey
Senior Lecturer
Department of Paediatric Dentistry
Glasgow Dental Hospital & School
378 Sauchiehall Street
Glasgow
G2 3JZ

Dear Dr Hosey

Trust Management Approval

LREC Ref: DENTAL 18 **R&D Project No: 03DN018**

Title: Hypodontia clinic database.

We are pleased to inform you that, based on the information provided, this project has received approval from our Finance Committee and the West Ethics Committee, and may now proceed. **We have recorded the Start date as 15/10/03 and the End date as** CONTINUOUS**, please let us know if there are any changes.**

Please note that Projects held on our Research Database are subject to scrutiny by the Chief Scientist Office (CSO) of the Scottish Executive. We are therefore required to keep this database up to date, and you will be requested to provide us with information on the progress, and ultimately the result of this research project. In addition, non-commercial projects are also entered onto the National Research Register. Details of this register are available from the West Research Office.

Thank you for your current and future collaboration.

Kind regards.

Yours sincerely

Judith Godden

Dr Judith Godden
Academic Research Co-ordinator

Appendix 2.

Information sheet for patients/volunteers for the Hypodontia Clinic database.

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THIS SHEET HAS BEEN APPROVED BY THE GGHB AREA DENTAL ETHICS COMMITTEE

INFORMATION SHEET FOR PATIENTS/VOLUNTEERS IN CLINICAL RESEARCH PROJECT
Brief Title of Project

Hypodontia clinic database.

Patient's Summary (Purpose of study, nature of procedure, discomfort and possible risks in terms which the patient or volunteer can understand).

The purpose of this project is to store the information that we normally collect about you on a database. This information will relate to your missing teeth (hypodontia)and will record details about your general and dental health. It will also include details of what we have found once we have examined your mouth and teeth, including moulds and the result of the x-ray examination.

Photographs will also be taken of you and your teeth, normally and these will also be stored on the database.

All of this information would normally be stored in your case notes.
We are asking your permission to include your details in this collection of information.

This will not involve any procedures or investigations that would not normally be carried out as part of your dental treatment.

The information stored about you will be anonymous, separate and secure.
We plan to use this information to see how well we manage patients with missing teeth.
We hope in the future to link this with how missing teeth can be inherited.

If you do not wish to have your clinical details stored in this way you need not agree to take part.
Even if you wish to have your details included now, you are free to change your mind later.

If you decide you do not want your details stored in this way,
this will not affect your dental treatment in any way.

Please address any concerns about this project to;
Dr Marie Therese Hosey,
Senior Lecturer in Paediatric Dentistry,
Glasgow Dental School,
378 Sauchiehall Street, Glasgow.

Tel.: 0141 211 9662

Appendix 3.

Consent form for patients/volunteers for the Hypodontia Clinic database.

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Format of Consent Form for Children

AREA DENTAL ETHICS COMMITTEE

FORM OF CONSENT FOR PATIENTS/VOLUNTEERS IN CLINICAL RESEARCH PROJECT

Title of Project :

Hypodontia clinic database.

By signing this form you give consent to your participation in the project whose title is at the top of this page. You should have been given a complete explanation of the project to your satisfaction and have been given the opportunity to ask questions. You should have been given a copy of the patient information sheet approved by the Area Dental Ethics Committee to read and to keep. Even though you have agreed to take part in the research procedures you may withdraw this consent at any time without the need to explain why and without any prejudice to your care.

Consent and parental permission:

I confirm that I have read the Patient Information Sheet and give my parental permission to the research procedures, the nature, purpose and possible consequences of which have been described to me by
.....ROSEMARY....BROAD.....
(PRINT researcher's name)

Parent's signature.....Date.....

Child's signature.....Date.....

Doctor / Dentists's signature.....Date.....

UNIVERSITY OF GLASGOW DENTAL SCHOOL
378 Sauchiehall Street, Glasgow G2 3JZ, UK
Switchboard: 0141-211 9600 *Fax:* 0141-331 2798

Appendix 4.

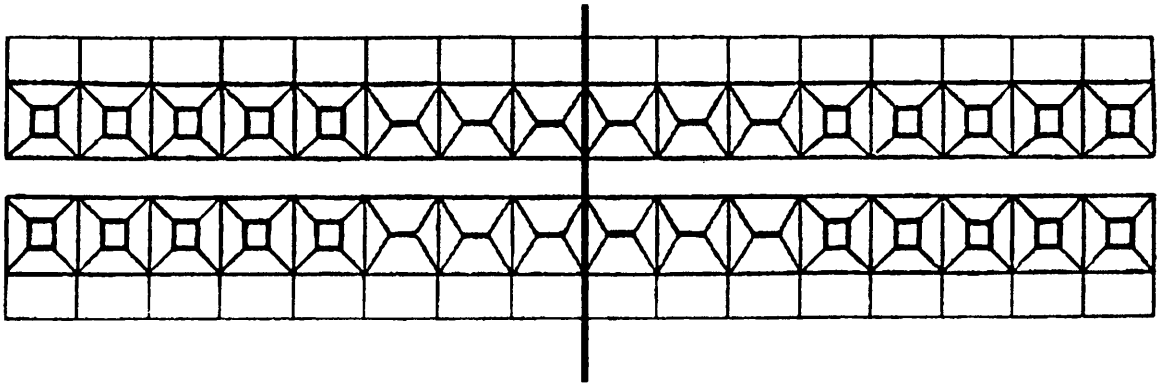
"Old" Hypodontia Clinic assessment form.

Surname	First Name(s)	Unit No.
Referred By *Please Circle* Ortho / paed / restorative	Initial Referring GDP/ CDS	Date
Examining Surgeon	Consultant(S)	Study model box number
Presenting Complaint		
History of Presenting Complaint		
PDH (including previous extractions/ ortho treatment)		
Family History		
Relevant Medical History		
Social History	Attitude	Habits

Clinical Examination

skeletal pattern	***antero posterior***	I / II / III	mild/ moderate / severe
	vertical	FMPA	average/ increased/ decreased
	Transverse (asymmetry)	Y / N comments	
TMJ	symptoms? Y / N comments:		
Soft tissues *extra oral*	***lips***	competent / incompetent	Draw lip line 
	lip line	high/ normal/ low	
intra oral	***periodontal tissues***	Gingivitis: Y/N comment	
		Plaque: Y/N Interproximal / smooth surface.	
	Other findings		

Dental Charting *- Enter location of teeth shaded in clear boxes, place identifiers of abnormal teeth above location boxes when appropriate*



Traumatised Teeth Y / N	
Tooth Tissue Loss Y / N *Distribution*	
Enamel Hypoplasia Y / N *Describe*	
Caries Risk Assessment – High/Average/low **Carious teeth** _____/_____ /	**Teeth of poor prognosis** _____/_____ /

Orthodontic Assessment

Please circle as appropriate

- **Lower labial segment** — **Inclination** — **Average/Proclined/Retroclined**
  Aligned/Crowded/Spaced — **Mild/Moderate/Severe**
  Rotated teeth
- **Upper labial segment** — **Inclination** — **Average/Proclined/Retroclined**
  Aligned/Crowded/Spaced — **Mild/Moderate/Severe**
  Rotated teeth

SPACE ANALYSIS

Mandible	$\overline{5}$/	$\overline{4}$/	$\overline{3}$/	$\overline{/3}$	$\overline{/4}$	$\overline{/5}$	Maxilla	5/	4/	3/	/3	/4	/5
	7	7	7	7	7	7		7	7	8	8	7	7
Space required	21			21			Space required	22			22		
Space available							Space available						
Space shortage/surplus (-) (+)							Space shortage/surplus (-) (+)						

- **Lower buccal segments** — **Aligned/Crowded/Spaced** — **Mild/Moderate/Severe**
- **Upper buccal segments** — **Aligned/Crowded/Spaced** — **Mild/Moderate/Severe**

- **Overbite** Average/Increased/Decreased Complete/Incomplete/Traumatic

  Anterior open bite .....mm

- **Incisor Relationship** I II Div 1 II Div 2 III
- **Centre lines** Upper-Right/Central/Left .....mm

  Lower-Right/Central/Left .....mm
- **Molar relationship** Right I / II / III ½ /full
  Left I / II / III ½ /full
- **Crossbites** Unilateral – Right/Left Displacement – Yes/No Teeth affected _____/_____
  Bilateral Displacement – Yes/No /

IOTN Score |

- **Occlusal pattern**

Report Of Other Investigations:

- **Sensibility Test** - **Tooth**
  (please indicate type) - **Response**

RADIOGRAPHS

- **OPT / occlusal** *Relevant findings*

  (periapicals if applicable)

- **Unerupted teeth** _____/_____ **MISSING TEETH** _____/_____

- **Lateral skull radiograph**

	Date				Date		
81 ± 3	SNA			27 ± 4	MMPA		
78 ± 3	SNB			109 ± 6	1 - MAX. P		
3 ± 2	ANB			93 ± 6	$\bar{1}$ - MP		
	Class			135 ±10	1 - $\bar{1}$		
				55±2%	Facial %		
				0-2 mm	$\bar{1}$ to APo		

Cephalogram Analysis

Summary of Findings

Aims Of Treatment

Treatment Plan

Records	**Dates:**				
• *Study casts*					
• *Photos*					
• *Radiographs*					

Signature(s)

Date

Risks and benefits of treatment explained Y / N

Consent Y / N

Letter to primary care provider?

Disposal

Review again on Hypodontia Clinic? Y / N

If yes when? .....

Appendix 5.

Ammended Hypodontia Clinic assessment form.

HYPODONTIA CLINIC PATIENT ASSESSMENT RECORD

Surname	First Name(s)	Unit No
Referred By Ortho/ Paed/ Restorative	Initially referred GDP/CDS	Date
DOB	Age on presentation hypodontia clinic	Study model box No
Examining Surgeon	Consultant(s)	
Presenting Complaint		
History of Presenting Complaint		
Relevant Medical History		
PDH (previous extractions/ ortho treatment)		
Family History		
Social History	Attitude	Habits

Clinical examination

Extraoral

Skeletal pattern	Antero posterior	I /II / III	Mild / moderate / severe
	Vertical	FMPA	Average / increased / decreased
	Transverse (asymmetry)	Y / N comments	
TMJ	Symptoms?	Y / N	
Soft tissues	Lips	Competent/ incompetent	Draw lip line
	Lip line	High / Normal/ Low	

Intra oral

Dental Charting

Teeth present

/
/

Teeth unerupted

/
/

Teeth Missing

/
/

Traumatised Teeth Y / N	
Tooth tissue loss Y / N	Mild / Moderate / Severe Distribution
Enamel Hypoplasia Y / N Describe	
Caries Risk Assessment – High/Average/low **Carious teeth** / /	**Plaque** Y / N **Gingivitis** Y / N **Teeth of poor prognosis** / /

Orthodontic Assessment

Incisor Relationship	**Overjet** .....mm	**Overbite** .....mm Average/ increased/ decreased Complete/incomplete/Traumatic
Centre lines / /	**Molar Relationship** **RHS** **LHS**	**Crossbite / Displacement** Teeth affected
Upper arch Spacing / Alignment		
Lower arch Spacing / Alignment		
Other features		
IOTN Score		

Report of Other Investigations:

Sensibility testing	Type	Tooth Response

Radiographs Relevant findings	Type

Aims of Treatment

.

.

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Treatment Plan

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Disposal

Department	Consultant in charge	Appointment made Y / N With Whom ?
Orthodontics		
Child Dental Health		
Conservation		
Oral Surgery		
Primary Care Provider		Inform the parent to arrange

Risks and benefit of treatment explained? Y / N
Letter to Primary Care Provider? Y / N
Review again on Hypodontia Clinic? Y / N

Signature(s)

Date **...../...../ 20.....**

Appendix 6.

Ethical approval for a prospective controlled study to evaluate self-esteem in adolescents with hypodontia from:

- West Glasgow Ethics Committee 2.
- North Glasgow University Hospital Research and Development.

North Glasgow University Hospitals
Division

NHS
Greater
Glasgow

West Glasgow Ethics Committee 2
Western Infirmary
Dumbarton Road
Glasgow
G11 6NT

Telephone: 0141 211 6238
Facsimile: 0141 211 1920

Email: andrea.torrie@northglasgow.scot.nhs.uk

23 December 2004

Dr M. T. Hosey
Senior Lecturer / Honorary Consultant
Glasgow Dental Hospital & School
378 Sauchiehall Street
Glasgow
Scotland, U.K.
G2 3JZ

Dear Dr Hosey

Full title of study: *Evaluation of how congenitally missing teeth affects feelings of self-esteem: a controlled trial.*
REC reference number: *04/S0709/88*
Protocol number:

The Research Ethics Committee reviewed the above application at the meeting held on 21 December 2004.

Ethical opinion

The Committee wished to thank yourself and Prof Bagg for attending the meeting to discuss the above study.

The Committee were generally in favour of the study but wondered what arrangements were in place to assist with patients who are identified as having low self-esteem. You confirmed input from the Consultant Psychologist who would be picking up on this issue where necessary.

The Patient Information Sheet should be amended to be "invitational".

The above minor amendments should come back to me for filing.

The members of the Committee present gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation.

Procedures for Research Ethics Committees in the UK.

04/S0709/88	**Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project,

Yours sincerely,

Andrea H Torrie
Administrator - West Ethics Committee

E-mail: andrea.torrie@northglasgow.scot.nhs.uk

Enclosures *List of names and professions of members who were present at the meeting*

Standard approval conditions

Site approval form (SF1)

The favourable opinion applies to the research sites listed on the attached form.

Conditions of approval

The favourable opinion is given provided that you comply with the conditions set out in the attached document. You are advised to study the conditions carefully.

Approved documents

The documents reviewed and approved at the meeting were:

Document Type:	Version:	Dated:	Date Received:
Application	Version 3 Jan 2004	29/11/2004	29/11/2004
Investigator CV		29/11/2004	29/11/2004
Protocol		29/11/2004	29/11/2004
Copy of Questionnaire		29/11/2004	29/11/2004
Participant Information Sheet		29/11/2004	29/11/2004
Participant Consent Form	Version 1	10/11/2004	29/11/2004

Management approval

The study should not commence at any NHS site until the local Principal Investigator has obtained final management approval from the R&D Department for the relevant NHS care organisation.

Membership of the Committee

The members of the Ethics Committee who were present at the meeting are listed on the attached sheet.

Notification of other bodies

The Committee Administrator will notify the research sponsor that the study has a favourable ethical opinion.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating

West Glasgow Ethics Committee 2

LIST OF SITES WITH A FAVOURABLE ETHICAL OPINION

For all studies requiring site-specific assessment, this form is issued by the main REC to the Chief Investigator and sponsor with the favourable opinion letter and following subsequent notifications from site assessors. For issue 2 onwards, all sites with a favourable opinion are listed, adding the new sites approved.

REC reference number:	04/S0709/88	**Issue number:**	1	**Date of issue:**	23 December 2004
Chief Investigator:	Dr M. T. Hosey				
Full title of study:	Evaluation of how congenitally missing teeth affects feelings of self-esteem: a controlled trial.				

This study was given a favourable ethical opinion by West Glasgow Ethics Committee 2 on 21 December 2004. The favourable opinion is extended to each of the sites listed below. The research may commence at each NHS site when management approval from the relevant NHS care organisation has been confirmed.

Principal Investigator	Post	Research site	Site assessor	Date of favourable opinion for this site	Notes [1]
Dr M. T. Hosey	Senior Lecturer / Honorary Consultant	Greater Glasgow Primary Care NHS Trust. North Glasgow University Hospital NHS Trust. Glasgow Dental Hospital & School, Paediatric Department. Community Dental Clinics, Glasgow.	NHS Greater Glasgow Primary Care Division (Community & Mental Health)	23/12/2004	

Approved by the Chair on behalf of the REC:

Andrea H Torrie ..... (Signature of ~~Chair~~/Administrator*)

(**delete as applicable)*

ANDREA H TORRIE (Name)

[1] *The notes column may be used by the main REC to record the early closure or withdrawal of a site (where notified by the Chief Investigator or sponsor), the suspension of termination of the favourable opinion for an individual site, or any other relevant development. The date should be recorded.*

Minutes of the meeting of the West Glasgow Ethics Committee 2 Research Ethics Committee held on 21 December 2004 at 09:00:00 in Western Infirmary, Catering Dept, Function Suite, Western Infirmary, G11 6NT

PRESENT:

Dr N Pace (Chairman)
Rev R Currie
Sister C Donald
Dr E Douglas
Dr S Langridge
Prof K R Lees
Mr J McHugh
Prof. B Parfitt
Dr R L Soutar
Mr K Wallace
Mrs J Wardlaw
Mrs A H Torrie

North Glasgow University Hospitals
Division

West Research Office
Administration Building
Ground Floor Room 9
Western Infirmary
Glasgow G11 6NT
Tel: 0141 211 6281

NHS
Greater
Glasgow

Our Ref JG/LR
Enquiries to Judith Godden
Direct Line 0141 211 1817
e-mail: *Judith.Godden@northglasgow.scot.nhs.uk*

26th Jan 2005

Dr M T Hosey
Senior Lecturer/Honorary Consultant
Glasgow Dental Hospital & School
378 Sauchiehall Street
Glasgow
G2 3JZ

Dear Dr Hosey

LREC Ref: 05/S0709/88 **R&D Project Ref:** WN05DN002
Project Title: Evaluation of how congenitally missing teeth affects feelings of self-esteem: a controlled trial.

We are pleased to inform you that, based on the information provided, this project has been granted overall Management Approval and may now proceed. This includes Finance, Pharmacy and a favourable Research Ethics Committee opinion.

Under Research Governance, we are required to hold a sponsor file containing the following documents: Protocol, Amendments and Ethics approval. While the study is ongoing you are responsible for updating us with all study amendments.

Further management approval will be required for amendments that increase patient numbers, increase or change the test procedures or bring about a change in pharmacy requirements. Please contact the R&D office if you wish to discuss any future amendments.

Thank you for your current and future collaboration.

Kind regards.

Yours sincerely

Judith Godden

Dr Judith Godden
Academic Research Co-ordinator

Appendix 7.

Information sheet for patients/parents /guardians invited to participate in this research project.

Information Sheet for Patients/ Parents/Guardians invited to participate in a Research Project.

Title of the project:

Evaluation of how congenitally missing teeth affects feelings of self-esteem.

My name is Rosemary Broad and I am involved in a project to find out how patients feel about their teeth. Some patients have missing teeth and I wish to find out if their feelings are different to those patients who have no missing teeth.

What is involved for the patient?

I would like to invite you to answer questions about your feelings about your teeth and how this affects how you feel generally.

There will be no extra visits required.

Your personal details will be kept confidential. No names will appear on any of the information.

The results will be published in a scientific journal and also presented at scientific meetings.

I would be grateful if you would agree to take part but then if you change your mind this will not affect your treatment in any way.

If you do agree to take part a consent form will be required to be signed by the patient and parent/guardian.

Thank you for taking the time to read this and if there is any further information you wish please contact:

Dr M.T. Hosey and Mrs R Broad.
0141 211 9665 (secretary)

Any queries about this project can be addressed to Dr M.T. Hosey:
Glasgow Dental Hospital & School
Paediatric Department
378 Sauchiehall Street
Glasgow, G2 3JZ.

Appendix 8.

Consent form for patients/parents/guardians for subjects to participate in this research project.

Version 1, 10.11.04

Consent Form for Patients/Parents/guardians in a Research Project.

Title of project:

Evaluation of how congenitally missing teeth affects feelings of self-esteem.

By signing this form you give consent to take part in the project about how patients feel about their teeth and how this affects them generally. You will have been spoken to about the project, given an information sheet, which has been approved by the Ethics Committee and also given the opportunity to ask questions.

You may change your mind about taking part in the project at any time and this will not affect your treatment in any way.

I, _____ (PRINT)

have been given an information sheet and give my consent to take part in the above project. I understand that my personal information will remain confidential and that I can withdraw from the project at any time without affecting my treatment.

Patient's signature

_____Date_____

Parent/guardian's signature

_____Date_____

Researcher's signature

_____Date_____

Appendix 9.

Hypodontia Clinic questionnaire.

Hosey-Bradnock questionnaire.

Hypodontia Clinic: Questionnaire. Patient Code

We would be grateful if you would read the questions below and answer them as best you can. For questions numbered 1 to 8 please circle the answer best suited to how you feel, an example is shown for you. For questions 9, 10 and 11 at the end we would like you to write in the space provided.

Thank you for your time and help.

Example:

How do you feel about watching football on the television?

hate (dislike) not bothered like love

Questions:

1. What is it about your teeth that you don't like?

Spaces / gaps shape of teeth teeth look uneven loose

2. How do you feel about how your teeth look?

hate dislike not bothered like happy

3. Do you feel self conscious or uncomfortable when you smile for photographs, at parties or meet new people?

Not at all a little fairly a lot extremely

4. Do you feel embarrassed about your teeth?

not at all a little fairly a lot extremely

5. Do you get teased at school because of your teeth?

Not at all sometimes fairly often often

6. How much does having missing teeth bother you?

Not at all a little quite a bit upsets me

7. What do you think about wearing false teeth (dentures)?

Happy to wear one would wear one but not happy about it would never wear one

8. What do you think about wearing braces?

Happy to wear one would wear one but not happy about it would never wear one

9. What would you want dental treatment to do for you?

10. What do you expect from this visit to the Hypodontia Clinic today?

11. What kind of dental treatment do you expect in the future?

Appendix 10.

Open questions of the Hosey-Bradnock questionnaire.

Appendix 10.

Open Question Responses from The Hosey-Bradnock questionnaire.

Question 9 **What would you want dental treatment to do for you?**
To make teeth look normal.
Implants. Secure teeth, dentures slack.
Help to make my teeth look better.
Make me look nice when I smile.
I would like to have natural teeth.
No gaps. No wonkey teeth.
Make my teeth look nice and even.
Fix the appearance of my teeth (straighten and no gaps).
Make my teeth better.
Sort the shape and positioning of my teeth.
Make my teeth look better.
To get implants when older and have the perfect smile.
To make my teeth look better.
Make it easy for me to get through it as possible, preferably without false teeth or dentures.
To straighten my teeth.
Fix my teeth.

Nothing.
To close gap in my front teeth.
Reduce gaps between teeth.
Make teeth healthy and nice looking.
Straighten all my teeth.
Fix my teeth.
Even out my teeth a little and keep the sizes the same.
Uncross my front teeth.
To do something so I have no gaps.
Help my teeth.
Have my teeth look nicer.
Improve how my front teeth look like.

Question 10 **What do you expect from this visit to the Hypodontia clinic today?**
A check up.
Good –got everything I wanted, everything explained to me.
They correct my teeth.
Maybe a brace.
To help decide what is going to happen next.
Do not know.
To know what's happening with my mouth, to get my teeth sorted.
Answers and further appointments to start fixing my teeth.
Not sure.
To help me through.
To find out what's happening with my teeth.
To plan what is going to happen in the future concerning my teeth.
To be examined and briefed.
I don't quite know yet.
Not recorded.
Not recorded.
Nothing.
Not recorded.
Find out how to make my teeth look better.
Not sure.
Not sure.

To find out what is to be planned with the condition of my teeth.
To be presented with choices of treatments that will help my teeth look better.
To have treatment to uncross my teeth.
I expect them to fill the gaps.
Good advice.
A check-up.
Information.

Question 11 **What kind of dental treatment do you expect in the future?**
Dentures put in.
Implants.
Braces and a false tooth.
Hopefully none.
I would like to have my spaces filled in with natural looking teeth.
Braces.
Implants.
Braces.
Don't know.
Here to find out.
Braces.
To get implants.
Not sure.
The good kind.
Not recorded.
Not recorded.
Don't know.
Not recorded.
Would like teeth straightened, evened and gaps reduced.
Painless.
Not sure.

Hasn't been discussed yet, hopefully something to improve my teeth.
Don't know.
Braces.
Braces and fill in spaces.
Not sure.
?
Don't know.

Appendix 11.

Presenting complaints of subjects with hypodontia.

Appendix 11.

Subjects Presenting Complaints
Unable to bite. Unhappy with aesthetics.
Nil c/o from patient. Referred from Orthodontist.
Nil c/o from patient.
Appearance of anterior teeth.
Spaces, canines too pointed, appearance.
Nil c/o at the moment, father concerned re the future.
Concerned about gaps in the front teeth.
Squint teeth. Missing teeth.
Not bothered.
Unhappy with spaces and increased centre line.
Unhappy with size of front teeth. Unhappy with gaps.
Unhappy with appearance of front teeth.
Dislikes appearance of front teeth. Likes denture but wishes something more permanent. Wants bridges.
Concerned about spaces which will be present once baby teeth fall out.
Not recorded.
Unhappy about appearance.
Nil c/o from patient.
Loose partial denture.
C/o spacing of front teeth. Wishes teeth the same size.
Dislikes spacing between upper central incisors.

Dislikes gaps. Hates all teeth.
Missing teeth.
Anxious for orthodontic treatment to start. Wishes teeth to look normal. Not concerned about appearance.
Concerned about gap between front teeth.
Upper denture loose.
Concerned about later on when baby teeth fall out.
Concerned about spaces at front teeth.
Nil c/o.
Nil c/o from patient.
Not concerned about teeth at the present.
Missing teeth bothers the parent.
Front teeth annoying, don't like gaps, teased at school.
Patient feels teeth are "all in a mess". Missing teeth.
Patient hates front teeth gaps and pointy bits.
Patient won't wear partial dentures.
Does not like appearance of lower front teeth.
Does not like appearance of front teeth. Worried about how teeth will look like in the future. Does not smile.
Not recorded.
Missing two teeth.
Does not like acrylic partial denture and spaces LHS. Mainly concerned about appearance of spaces at the top. CoCr did not fit.
At present not bothered about appearance, although overlapping upper central

incisors felt getting worse. Diastema in lower central region.
Appearance.
Not recorded.
Currently wearing twin block appliance therapy. Concerned about appearance now wearing appliance.
Nil.
Nil mentioned by patient but father reports that he is being teased at school "gappy". Anterior maxillary aesthetics.
Unhappy with appearance and spacing.
Aware of spaced upper incisors.
Crooked teeth, thinks needs a brace.
Not bothered about teeth but mother is.
Not bothered at the moment but concerned about the future.
Spacing on upper front teeth.
Spacing and what happens when my baby teeth fall out.
Missing teeth.
Chipped tooth.
Gap UL3 region, as ULC had exfoliated. Worried about URC region once it exfoliates.
Patient not bothered. Mum very concerned.
Missing teeth and wearing a lower partial denture.
Big front teeth stick out. Missing back teeth.
Spaces upper and lower arch but pointed to upper anteriors specifically.
Missing teeth, spaces between teeth. Does not wish to wear dentures at any point.

Might like them straighter.
Missing teeth and teeth different sizes.
Spaces between teeth.
Front teeth crossing over, don't like.
A little bothered about gaps.
Chipping of UL1.
Teeth not in the right place and there are gaps.
No complaint but feels strange to chew.
Does not like gaps and angle of UL1 "horrible teeth"
No complaint from patient but Mum concerned about what will happen when she grows.

Appendix 12.

Culture-Free Self-Esteem Inventory-Third Edition.

Adolescent Student Response Form.

CFSEI-3

Culture-Free Self-Esteem Inventories–Third Edition

Adolescent Student Response Form

Name _____ Female ☐ Male ☐

School _____ Date of Testing _____

Grade _____ Date of Birth _____

Instructions: Read each statement and mark the circle or square to respond yes or no.

Item	Statement	Yes	No	Score
1.	Do you have only a few friends?	○	○	
2.	Are you happy most of the time?	☐	☐	
3.	Are you comfortable telling your parents about your problems?	☐	☐	
4.	Can you do things as well as others?	☐	☐	
5.	Do you like everyone you know?	☐	☐	
6.	Do you spend most of your free time alone?	○	○	
7.	Do you trust your family?	☐	☐	
8.	Do most people like you?	☐	☐	
9.	Do you often feel like quitting school?	○	○	
10.	Have you ever taken anything that didn't belong to you?	○	○	
11.	Are you as intelligent as most people?	☐	☐	
12.	Do you argue often with your family?	○	○	
13.	Do you feel you are as important as most people?	☐	☐	
14.	Are you easily depressed?	○	○	
15.	Are you satisfied with your schoolwork?	☐	☐	
16.	Do you feel like you have enough freedom at home?	☐	☐	
17.	Would you change many things about yourself if you could?	○	○	
18.	Do you always tell the truth?	☐	☐	
19.	Do you have a good relationship with your mother?	☐	☐	
20.	Are you as nice looking as most people?	☐	☐	
21.	Do you usually quit when your schoolwork is too hard?	○	○	
22.	Are you usually tense or anxious?	○	○	
23.	Do you gossip at times?	○	○	
24.	Do you often feel that you are no good at all?	○	○	
25.	Are you as strong and healthy as most people are?	☐	☐	
26.	Do you enjoy learning?	☐	☐	
27.	Do you have a good relationship with your father?	☐	☐	
28.	Is it difficult for you to express your views and feelings?	○	○	
29.	Do you ever get angry?	○	○	

2 3 4 5 06 05 04 03 02

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Item	Statement	Yes	No	Score
30.	Do you often feel ashamed of yourself?	○	○	
31.	Are you pretty good about doing homework on time?	□	□	
32.	Are other people generally more successful than you are?	○	○	
33.	Are you doing as well in school as you would like to do?	□	□	
34.	Do you feel uneasy most of the time without knowing why?	○	○	
35.	Do you feel that you are as happy as others are?	□	□	
36.	Are you ever shy?	○	○	
37.	Are you a failure?	○	○	
38.	Are you popular with other people your age?	□	□	
39.	Is it hard for you to meet new people?	○	○	
40.	Do you ever lie?	○	○	
41.	Are you often upset about something?	○	○	
42.	Do most people respect your views?	□	□	
43.	Are people your age smarter than you are?	○	○	
44.	Are you uncomfortable in groups of people?	○	○	
45.	Do you worry more than most people do?	○	○	
46.	Are you as happy as most people?	□	□	
47.	Do the people in your family have quick tempers?	○	○	
48.	Are you ever sad?	○	○	
49.	Is it hard for you to find the energy to do things?	○	○	
50.	Do you feel as though your friends have a lot of confidence in you?	□	□	
51.	Do you feel that you are not good enough?	○	○	
52.	Do your parents understand how you feel?	□	□	
53.	Are you doing the best schoolwork that you can?	□	□	
54.	Do people like your ideas?	□	□	
55.	Do your parents think you are a failure?	○	○	
56.	Is it hard for you to give yourself credit when you do a good job?	○	○	
57.	Do you have trouble talking to other people?	○	○	
58.	Are you a failure at school?	○	○	
59.	Do your parents love you?	□	□	
60.	Are you proud of your schoolwork?	□	□	
61.	Do you often get upset when you are at home?	○	○	
62.	Are you a hard worker at school?	□	□	
63.	Do you find it hard to make up your mind and stick to it?	○	○	
64.	Do you like yourself very much?	□	□	
65.	Do you have friends you can confide in?	□	□	
66.	In school, do you do as little work as you can get by with?	○	○	
67.	Do you feel as though your family trusts you?	□	□	

Appendix 13.

Culture-Free Self-Esteem Inventory-Third Edition.

Adolescent (Ages 13-18).

Profile/Scoring Form.

CFSEI–3

Culture-Free Self-Esteem Inventories–Third Edition

Adolescent (Ages 13–18)

Profile/Scoring Form

Section I. Identifying Information

Name _____

School _____

Grade _____ Female ☐ Male ☐

Examiner _____

Examiner's Title _____

	Year	Month	Day
Date Tested	_____	_____	_____
Date of Birth	_____	_____	_____
Test Age	_____	_____	_____

Section II. Record of Scores

Subscales	Raw Scores	Standard Scores	%iles	Descriptive Ratings (from Table 3.1 in manual)
Academic (A)	_____	☐	_____	_____
General (G)	_____	☐	_____	_____
Parental/Home (P/H)	_____	☐	_____	_____
Social (S)	_____	☐	_____	_____
Personal (P)	_____	☐	_____	_____
Sum of Subscales		_____		
Global Self-Esteem Quotient		◯	_____	_____

Defensiveness (D) Score _____ out of 8 ←

The recommended cutoff score is 4 out of 8. This score indicates the extent to which the defensiveness of the child may diminish the validity of the Quotient.

Section III. Profile of Scores

Standard Scores	Academic (A)	General (G)	Parental/Home (P/H)	Social (S)	Personal (P)	Standard Scores	Quotient	Global Self-Esteem
20	•	•	•	•	•	20	150	•
19	•	•	•	•	•	19	145	•
18	•	•	•	•	•	18	140	•
17	•	•	•	•	•	17	135	•
16	•	•	•	•	•	16	130	•
15	•	•	•	•	•	15	125	•
14	•	•	•	•	•	14	120	•
13	•	•	•	•	•	13	115	•
12	•	•	•	•	•	12	110	•
11	•	•	•	•	•	11	105	•
10	—	—	—	—	—	10	100	—
9	•	•	•	•	•	9	95	•
8	•	•	•	•	•	8	90	•
7	•	•	•	•	•	7	85	•
6	•	•	•	•	•	6	80	•
5	•	•	•	•	•	5	75	•
4	•	•	•	•	•	4	70	•
3	•	•	•	•	•	3	65	•
2	•	•	•	•	•	2	60	•
1	•	•	•	•	•	1	55	•

Section IV. Examination Conditions

Who referred the child? _____

What was the reason for referral? _____

_____

Place tested _____

_____

	Interfering				Not Interfering
Noise level	1	2	3	4	5
Interruptions	1	2	3	4	5
Distractions	1	2	3	4	5
Light	1	2	3	4	5
Temperature	1	2	3	4	5
Understanding	1	2	3	4	5
Energy level	1	2	3	4	5
Attitude	1	2	3	4	5
Rapport w/ examiner	1	2	3	4	5
Any other _____	1	2	3	4	5

Notes and other considerations: _____

_____

_____

_____

_____

1 2 3 4 5 06 05 04 03 02

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Section V. Score Summary

Item	Score	A	G	P/H	S	P	D
1.	___				___		
2.	___		___				
3.	___			___			
4.	___		___				
5.	___						___
6.	___				___		
7.	___			___			
8.	___				___		
9.	___		___				
10.	___						___
11.	___		___				
12.	___			___			
13.	___		___				
14.	___					___	
15.	___	___					
16.	___			___			
17.	___					___	
18.	___						___
19.	___			___			
20.	___		___				
21.	___	___					
22.	___					___	
23.	___						___
24.	___					___	
25.	___		___				
26.	___	___					
27.	___			___			
28.	___				___		
29.	___						___
30.	___					___	
31.	___	___					
32.	___		___				
33.	___	___					
34.	___					___	

Item	Score	A	G	P/H	S	P	D
35.	___					___	
36.	___						___
37.	___		___				
38.	___				___		
39.	___				___		
40.	___						___
41.	___					___	
42.	___				___		
43.	___		___				
44.	___				___		
45.	___					___	
46.	___					___	
47.	___			___			
48.	___						___
49.	___					___	
50.	___				___		
51.	___					___	
52.	___			___			
53.	___	___					
54.	___				___		
55.	___			___			
56.	___					___	
57.	___				___		
58.	___	___					
59.	___			___			
60.	___	___					
61.	___			___			
62.	___	___					
63.	___					___	
64.	___		___				
65.	___				___		
66.	___	___					
67.	___			___			

	A	G	P/H	S	P	D
Raw Scores	☐	☐	☐	☐	☐	☐

Section VI. Interpretation and Recommendations

Appendix 14.

Culture-Free Self-Esteem Inventory-Third Edition.

Adolescent Student Response Form.

Highlighting eight questions incorporating the "lie detector".

CFSEI-3

Culture-Free Self-Esteem Inventories–Third Edition

Adolescent

Student Response Form

Name _____ Female ☐ Male ☐

School _____ Date of Testing _____

Grade _____ Date of Birth _____

Instructions: Read each statement and mark the circle or square to respond yes or no.

Item	Statement	Yes	No	Score
1.	Do you have only a few friends?	○	○	
2.	Are you happy most of the time?	□	□	
3.	Are you comfortable telling your parents about your problems?	□	□	
4.	Can you do things as well as others?	□	□	
5.	Do you like everyone you know?	□	□	
6.	Do you spend most of your free time alone?	○	○	
7.	Do you trust your family?	□	□	
8.	Do most people like you?	□	□	
9.	Do you often feel like quitting school?	○	○	
10.	Have you ever taken anything that didn't belong to you?	○	○	
11.	Are you as intelligent as most people?	□	□	
12.	Do you argue often with your family?	○	○	
13.	Do you feel you are as important as most people?	□	□	
14.	Are you easily depressed?	○	○	
15.	Are you satisfied with your schoolwork?	□	□	
16.	Do you feel like you have enough freedom at home?	□	□	
17.	Would you change many things about yourself if you could?	○	○	
18.	Do you always tell the truth?	□	□	
19.	Do you have a good relationship with your mother?	□	□	
20.	Are you as nice looking as most people?	□	□	
21.	Do you usually quit when your schoolwork is too hard?	○	○	
22.	Are you usually tense or anxious?	○	○	
23.	Do you gossip at times?	○	○	
24.	Do you often feel that you are no good at all?	○	○	
25.	Are you as strong and healthy as most people are?	□	□	
26.	Do you enjoy learning?	□	□	
27.	Do you have a good relationship with your father?	□	□	
28.	Is it difficult for you to express your views and feelings?	○	○	
29.	Do you ever get angry?	○	○	

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30.	Do you often feel ashamed of yourself?	○	○
31.	Are you pretty good about doing homework on time?	□	□
32.	Are other people generally more successful than you are?	○	○
33.	Are you doing as well in school as you would like to do?	□	□
34.	Do you feel uneasy most of the time without knowing why?	○	○
35.	Do you feel that you are as happy as others are?	□	□
36.	Are you ever shy?	○	○
37.	Are you a failure?	○	○
38.	Are you popular with other people your age?	□	□
39.	Is it hard for you to meet new people?	○	○
40.	Do you ever lie?	○	○
41.	Are you often upset about something?	○	○
42.	Do most people respect your views?	□	□
43.	Are people your age smarter than you are?	○	○
44.	Are you uncomfortable in groups of people?	○	○
45.	Do you worry more than most people do?	○	○
46.	Are you as happy as most people?	□	□
47.	Do the people in your family have quick tempers?	○	○
48.	Are you ever sad?	○	○
49.	Is it hard for you to find the energy to do things?	○	○
50.	Do you feel as though your friends have a lot of confidence in you?	□	□
51.	Do you feel that you are not good enough?	○	○
52.	Do your parents understand how you feel?	□	□
53.	Are you doing the best schoolwork that you can?	□	□
54.	Do people like your ideas?	□	□
55.	Do your parents think you are a failure?	○	○
56.	Is it hard for you to give yourself credit when you do a good job?	○	○
57.	Do you have trouble talking to other people?	○	○
58.	Are you a failure at school?	○	○
59.	Do your parents love you?	□	□
60.	Are you proud of your schoolwork?	□	□
61.	Do you often get upset when you are at home?	○	○
62.	Are you a hard worker at school?	□	□
63.	Do you find it hard to make up your mind and stick to it?	○	○
64.	Do you like yourself very much?	□	□
65.	Do you have friends you can confide in?	□	□
66.	In school, do you do as little work as you can get by with?	○	○
67.	Do you feel as though your family trusts you?	□	□

Appendix 15.

Culture-Free Self-Esteem Inventory-Third Edition.

Adolescent (Ages 13-18).

Profile/Scoring Form.

Showing the Defensive Score cut off point.

CFSEI–3

Culture-Free Self-Esteem Inventories–Third Edition

Adolescent (Ages 13–18)

Profile/Scoring Form

Section I. Identifying Information

Name _____

School _____

Grade _____ Female ☐ Male ☐

Examiner _____

Examiner's Title _____

	Year	Month	Day
Date Tested	_____	_____	_____
Date of Birth	_____	_____	_____
Test Age	_____	_____	_____

Section II. Record of Scores

Subscales	Raw Scores	Standard Scores	%iles	Descriptive Ratings (from Table 3.1 in manual)
Academic (A)	_____	☐	_____	_____
General (G)	_____	☐	_____	_____
Parental/Home (P/H)	_____	☐	_____	_____
Social (S)	_____	☐	_____	_____
Personal (P)	_____	☐	_____	_____
Sum of Subscales		_____		
Global Self-Esteem Quotient		◯	_____	_____

Defensiveness (D) Score _____ out of 8 ←

The recommended cutoff score is 4 out of 8. This score indicates the extent to which the defensiveness of the child may diminish the validity of the Quotient.

Section III. Profile of Scores

Standard Scores	Academic (A)	General (G)	Parental/Home (P/H)	Social (S)	Personal (P)	Standard Scores	Quotient	Global Self-Esteem
20	•	•	•	•	•	20	150	•
19	•	•	•	•	•	19	145	•
18	•	•	•	•	•	18	140	•
17	•	•	•	•	•	17	135	•
16	•	•	•	•	•	16	130	•
15	•	•	•	•	•	15	125	•
14	•	•	•	•	•	14	120	•
13	•	•	•	•	•	13	115	•
12	•	•	•	•	•	12	110	•
11	•	•	•	•	•	11	105	•
10	—	—	—	—	—	10	100	—
9	•	•	•	•	•	9	95	•
8	•	•	•	•	•	8	90	•
7	•	•	•	•	•	7	85	•
6	•	•	•	•	•	6	80	•
5	•	•	•	•	•	5	75	•
4	•	•	•	•	•	4	70	•
3	•	•	•	•	•	3	65	•
2	•	•	•	•	•	2	60	•
1	•	•	•	•	•	1	55	•

Section IV. Examination Conditions

Who referred the child? _____

What was the reason for referral? _____

Place tested _____

	Interfering				Not Interfering
Noise level	1	2	3	4	5
Interruptions	1	2	3	4	5
Distractions	1	2	3	4	5
Light	1	2	3	4	5
Temperature	1	2	3	4	5
Understanding	1	2	3	4	5
Energy level	1	2	3	4	5
Attitude	1	2	3	4	5
Rapport w/ examiner	1	2	3	4	5
Any other _____	1	2	3	4	5

Notes and other considerations: _____

1 2 3 4 5 06 05 04 03 02

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Section V. Score Summary

Item	Score	A	G	P/H	S	P	D
1.	___				___		
2.	___		___				
3.	___			___			
4.	___		___				
5.	___						___
6.	___				___		
7.	___			___			
8.	___				___		
9.	___		___				
10.	___						___
11.	___		___				
12.	___			___			
13.	___		___				
14.	___					___	
15.	___	___					
16.	___			___			
17.	___					___	
18.	___						___
19.	___			___			
20.	___		___				
21.	___	___					
22.	___					___	
23.	___						___
24.	___					___	
25.	___		___				
26.	___	___					
27.	___			___			
28.	___				___		
29.	___						___
30.	___					___	
31.	___	___					
32.	___		___				
33.	___	___					
34.	___					___	

Item	Score	A	G	P/H	S	P	D
35.	___					___	
36.	___						___
37.	___		___				
38.	___				___		
39.	___				___		
40.	___						___
41.	___					___	
42.	___				___		
43.	___		___				
44.	___				___		
45.	___					___	
46.	___					___	
47.	___			___			
48.	___						___
49.	___					___	
50.	___				___		
51.	___					___	
52.	___			___			
53.	___	___					
54.	___				___		
55.	___			___			
56.	___					___	
57.	___				___		
58.	___	___					
59.	___			___			
60.	___	___					
61.	___			___			
62.	___	___					
63.	___					___	
64.	___		___				
65.	___				___		
66.	___	___					
67.	___			___			

	A	G	P/H	S	P	D
Raw Scores	☐	☐	☐	☐	☐	☐

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Abstract

R.D. Broad, M.T. Hosey. (2006). An evaluation of self-esteem in adolescents with hypodontia. Book of abstracts, 8th Congress of the European Academy of Paediatric Dentistry. Abstract 6-15. Amsterdam 2006.

Aim.

To report on the self-esteem of patients with hypodontia compared to non-hypodontia controls.

Method.

Prospective ethically approved, controlled study. The hypodontia group was 24 patients aged 12-17 years, 8 males, randomly selected from the multi-disciplinary Hypodontia Clinic. The control group was a random selection of 36 patients aged 12-18 years, 17 males, attending a local Community Clinic. All recruits were invited to complete the Culture-Free Self-Esteem Inventory consisting of 60 questions with a resultant numerical Global Self-Esteem Quotient {J Battle. Culture-Free Self-Esteem Inventories 3rd Edition, 2002, Texas, USA}. This includes subscales:- academic; general; parental / home; social and personal attributes giving a numerical Standard Score together with a "lie detector" test.

Results.

The mean Global Self-Esteem Quotient for the hypodontia group and the control group were 109.9 [range 83 to 122] and 108.8 [range 77 to 123] respectively. In respect to the subscales, the Standard Scores for the hypodontia compared to the controls were as follows: (1) hypodontia group: academic-mean 11.8 [range 5 to 13]; general- mean 10.3 [range 2 to 13]; parental/home-mean 12.2 [range 6 to 14];

social- mean 11.7 [range 6 to 13] and personal-mean 11.5 [range 5 to 14]; (2) control group: academic-mean 11.5 [range 5 to 13]; general-mean 9.9 [range 2-13]; parental/home-mean 12.4 [range 9 to 14]; social- mean 11.5 [range 3 to 13] and personal-mean 11.3 [range 7 to 14]. Twenty-nine percent of the hypodontia group were found to have "lied" compared to 38.9% of controls. There was no statistical difference between the hypodontia and control groups in respect to self-esteem.

Conclusion.

Patients with hypodontia did not have lowered self-esteem.